

Open Research Online

The Open University's repository of research publications and other research outputs

Sexuality, Relationships and Reproductive Choices in Young Adults with Life-Limiting and/or Life-Threatening Conditions

Thesis

How to cite:

Blackburn, M. C. (2019). Sexuality, Relationships and Reproductive Choices in Young Adults with Life-Limiting and/or Life-Threatening Conditions. PhD thesis The Open University.

For guidance on citations see [FAQs](#).

© 2018 The Author



<https://creativecommons.org/licenses/by-nc-nd/4.0/>

Version: Version of Record

Link(s) to article on publisher's website:

<http://dx.doi.org/doi:10.21954/ou.ro.0000f193>

Copyright and Moral Rights for the articles on this site are retained by the individual authors and/or other copyright owners. For more information on Open Research Online's data [policy](#) on reuse of materials please consult the policies page.

oro.open.ac.uk

Sexuality, relationships and reproductive choices in young adults
with life-limiting and/or life-threatening conditions

Madeleine Clare Blackburn

MSc, Postgraduate Diploma Law, Postgraduate Diploma Legal
Practice

Thesis presented for the degree of Doctorate in Philosophy

The Open University

Faculty of Wellbeing, Education and Language Studies

April 2018

Funded by The Open University



Frontispiece: Brighton Social Inclusion Parade, 2016 © Maddie Blackburn, 2016.

Abstract

This thesis details an empirical exploration of how young adults with life-limiting and/or life-threatening conditions talk about sexuality, intimacy, relationships and reproductive choices; what these mean, the difficulties they encounter along an uncertain life course, their relationship experiences, and what would be helpful to assist and make their lives more meaningful.

The research centres on 13 young adults aged over 16 years, featuring some, but not all, life-limiting or life-threatening conditions. These include cystic fibrosis, duchenne muscular dystrophy, other progressive neuromuscular and rare conditions, certain cancers and other genetic conditions. The research also explores the views and contributions of family supporters: two partners, ten parents and ten care practitioners, seeking their views in order to advance the young adult's sexual and relationship fulfilment. Until recently, surviving into adulthood with a life-limiting or life-threatening condition was unusual. Young adults with these conditions are a growing population and it is timely to explore their personal choices and considerations around sexuality, intimate relationships, and reproductive choices. The research used a qualitative approach, underpinned by a life course theoretical perspective, in conjunction with the illness experience and intersectionality with sexuality on an uncertain life course, to make sense of the participants' varied and personal contributions. What emerged was a picture of the transitions which occur from childhood to adulthood, from comparative wellness to progressive illness, uncertainty about their life course and importantly, their sexuality.

The findings suggest that the voices of this group are insufficiently heard and that there is a gap in the provision of information with respect to their transition from childhood to adulthood, particularly in relation to their sexuality and reproductive choices. As this group strives for independence and sexual citizenship, efforts need to be congruent with and grounded in the views of those most personally involved, the young adults themselves.

Dedication

This thesis is dedicated to all the young adults with life-limiting or life-threatening conditions who have a life even when it presents uncertainties.

Acknowledgements

First, I would like to thank all the participants who took part in this research. They generously gave their time, and shared intimate stories about their lives. Without their thoughtful and important contributions, this research would never have happened; second, The Open University for awarding me a studentship to carry out this research. Particular thanks go to my supervisors, Drs Sarah Earle and Carol Komaromy, for their unending patience and support, encouragement and excellent guidance throughout the ‘ups and downs’ of my doctoral journey.

My thanks also go to Dr Lindsay O Dell and Dr Duncan Banks, The Open University, Dr Lorna Fraser, Paediatric Epidemiologist, Martin House Research Centre, and Dame Elizabeth Fradd DBE.

Third, to doctoral colleagues and members of The Open University Sexuality Alliance, in particular, Alison Cooke, Lucy Watts MBE and Molly the Assistant Dog.

Fourth, Together for Short Lives, Hospice UK and other organisations in England for helping to source participants.

Thank you to my husband, Paul, who provided unconditional love and support when the ‘going got tough’ and for his technical wisdom in helping me to merge and format this thesis. Finally, to Lulu, our Siamese cat for ‘pawing’ the odd line of gobbledy-gook, as she skidded, uninvited, along the computer keyboard.

Table of Contents

Abstract	iii
Dedication	iv
Acknowledgements	v
Tables	xiv
Figures	xv
Chapter 1 An uncertain journey: Transitions	1
Introduction	1
1.1 Aims and rationale	5
1.2 Research context	7
Why now?	8
1.3 Personal transition: Why me?	9
1.4 Transitions in determining terminological usage	11
1.5 Transition and emerging adulthood	13
1.6 Sex(uality) as a shorthand	15
1.7 Transitions: The history and philosophy of palliative care	16
1.8 Transition: The emergence of palliative care for young adults	21
1.9 Classification and aetiology of LLTCs	23
1.10 Structure of this thesis	24
1.11 Conclusion	26
Chapter 2 Literature review	28
Introduction	28
2.1 Sexuality, sexual agency and intimate citizenship	30

	Sexuality	30
	Sexual Agency	31
	Intimate and Sexual Citizenship	33
2.2	Emerging adulthood	36
2.3	Empirical research: sexuality and disability	41
	Sexuality and people with life-limiting conditions	42
	Sexuality and people with life-threatening conditions	44
	Sex and relationship education (SRE)	47
	Balancing the rights to sexual expression with ‘protection from harm’	50
	Reproductive opportunities, choices and loss	52
2.4	Life course theoretical perspectives	53
	Life course: Principles	59
	Time, place and life-span development	59
	Timing	60
	Linked lives	60
	Transition points	61
	Liminality	63
	Uncertainty	66
	Biographical disruption	68
2.5	Conclusion	70
Chapter 3	Methodology and Methods	72
	Introduction	72
3.1	Methodology: Choosing a pragmatic qualitative approach	73

	Qualitative methodology	74
	Pragmatic	74
	The Life course perspective	75
	Answering the research questions	76
3.2	Methods and rationale	77
	Choice of method: Face to Face Interview	78
	Reasons for not choosing a questionnaire	79
	Ethics	79
	Gender	80
	Recruitment	82
	Research sample	84
	Voluntarism	85
	Consent and Confidentiality	87
	Research briefings	88
	Pilot study focus groups	88
	Main Study: Fieldwork overview and process	90
	Purposive selection of participants	90
	Main Study Interviews	90
	Pseudonyms and preferences	92
	Building rapport and other considerations	94
	Data analysis	95
3.3	Specific ethical and methodological challenges	100
	Gatekeeping	100

	Sensitive research	103
	Issues related to the research topic and the participants	105
	Issues related to the researcher	106
	Posthumous consent	107
3.4	Personal Reflection	107
	The Participants	109
3.5	Conclusion	110
Chapter 4	Uncertainty along the life course	111
	Introduction	111
4.1	The pivotal moment of diagnosis	113
4.2	Diagnosis at birth or in early childhood	114
4.3	Those diagnosed in later childhood or adolescence	121
4.4	Those with a recurring life-limiting and/or life-threatening conditions	130
4.5	Those with an unnamed or rare life-limiting and/or life-threatening condition	134
4.6	Discussion	138
	Timing of the diagnosis	139
	What's in the name?	140
	Fears of recurrence	141
4.7	Conclusion	143
Chapter 5	Sex and relationship education	144
	Introduction	144
5.1	Formal and informal sex and relationship education	145
5.2	Where, when and how	150

	Curriculum content and preferences	153
	Views about sex and relationship education	154
5.3	Informal opportunities to learn about sex(uality)	157
	Opportunities to access specialist sex and relationship education	161
5.4	Discussion	164
	Access to formal and informal specialist education	165
5.5	Conclusion	171
Chapter 6	The meanings and experiences of sex(uality)	172
	Introduction	172
6.1	The meanings of sex(uality)	172
	Friendships	173
	Relationships	181
	Intimacy	185
	Sex	187
6.2	Relationship experiences: sex(uality)	190
	Experiences of sex(uality): those in relationships	190
	Relationship experiences: those who had 'had sex'	195
	Those who had not had sex	196
6.3	Sexual satisfaction and relief	198
	Internet porn	200
	Sex workers and lap dancing	201
6.4	Barriers to relationships	202
	Presence of a life-limiting and/or life-threatening condition	203

	The impact on partners	206
	Choices and protection from harm	208
6.5	Discussion	209
	The meanings of sex(uality)	209
	Relationship opportunities	211
	Experiences of sex(uality)	211
	The barriers	213
6.6	Conclusion	215
Chapter 7	Uncertainty, reproductive hopes, choices and loss	216
	Introduction	216
7.1	Uncertainties about puberty and contraception	218
	Summary	230
7.2	Genetics and fertility	231
	Summary	242
7.3	Reproductive opportunities and the future	242
	Summary	248
7.4	Discussion	249
	Genetics	250
7.5	Conclusion	253
Chapter 8	Conclusions and recommendations	255
	Introduction	255
8.1	Aspects of life course	256
	Transition, liminality, and emergence	256

	Uncertainty and transition and turning points	257
	Uncertainty and biographical disruption	258
	Pivotal moment of diagnosis and its impact on sexuality	260
8.2	Research questions: key findings	261
	Relationship experiences	262
	Reproduction	263
8.3	The meta-learning	267
	Posthumous consent	267
	Data management and repositories	269
8.4	Limitations of this study	272
8.5	Recommendations for research, policy and practice	274
	Empirical findings: Implications for policy, practice and education	276
8.6	Final reflections	279
	Reflections on my doctoral journey	279
	Reflections on the findings	283
	Reflections about the co-researcher model	284
8.7	Conclusion	287
	References	288
	Appendix A: Abbreviations and definitions	333
	Appendix B: An overview of current UK Law (at the time of writing)	354
	Appendix C: Literature review Strategy	362
	Appendix D: Research project ethics approval	363
	Appendix E: Young adults' information sheet	364

Appendix F: Parents' and partners' information sheet	369
Appendix G: Care practitioners' information sheet	374
Appendix H: Consent forms	379
Appendix I: Life-limiting and/or life-threatening conditions relevant to this research	385
Appendix J: Research prompts	390
Appendix K: An excerpt of an interview transcript, illustrating how it was analysed.	392

Tables	Page
Table 1: The four main categories of life-limiting and life-threatening conditions.	23
Table 2: All participants and classifications.	96
Table 3: Process of Data Analysis in four stages.	99
Table 4: Participants with a known diagnosis of a LLTC at birth or in early childhood.	115
Table 5: Participants who knew, or were told, their LLTC diagnosis during adolescence.	122
Table 6: A typology of reproductive loss.	217

Figures		Page
Frontispiece:	Brighton Social Inclusion Parade, 2016.	ii
Figure 1:	Sex(uality) used as a shorthand for the research.	15
Figure 2:	My conceptual model for applying Elder's 'Life course Perspective' to the research design and data analysis.	57
Figure 3:	Stages of the research process and each of the organisations involved in the research, mapped onto a timeline, including those who eventually declined.	83
Figure 4:	The impact of the study and recommendations for future research, for practice and for education.	274
Figure 5:	A dissemination plan, outlining intentions including publications, future research and suggestions for education.	278

Chapter 1 An uncertain journey: Transitions

Introduction

'In my dreams I'm always able-bodied. Either I don't admit to myself that I'm disabled, or I feel that by will alone I can overcome it.' (Professor Stephen Hawking, reproduced in *The Times*, 14 March 2018)

Until recently, children and young people with life-limiting and/or life-threatening conditions (LLTCs) were not expected to live into adulthood (Beresford and Stuttard, 2014; Beresford, 2004). Life-limiting and/or life-threatening conditions are defined as:

'diseases with no reasonable hope of cure that will ultimately be fatal.' (Fraser et al., 2012, p. 924)

A key feature of children and young people born with or acquiring life-limiting and /or life-threatening conditions in childhood or adolescence is the diagnostic variation within the general population. Fewer children and young people with life-limiting and/or life-threatening (from hereon referred to as LLTCs) conditions today are less likely to have a malignant condition than their adult counterparts, and many children and young people may have one or more LLTCs, including congenital anomalies, chromosomal disorders and neuromuscular degenerative conditions (Hain et al, 2013b; Goldman, Hain and Lieben, 2012; Hain and Devins, 2011).

Due to advancements in medical technologies and clinical treatments, a new population of people with LLTCs are now living beyond original expectations into early adulthood and sometimes beyond (Beresford and Stuttard, 2014). There are approximately 49,000 people with a LLTC living in England (Fraser, 2012). Survival is most marked in 16 to 19 year olds and research has reported that the percentage of

children and young people up to 19 years with life-limiting conditions in England has more than doubled previous estimates, whilst percentages in Scotland, Wales and Northern Ireland were also reported to be higher than previous recordings (Fraser et al, 2015; 2013). There are around 13,000 people in the 18–25 year age group who are living with a LLTC condition in England (Fraser et al, 2013). The number of people with a LLTC globally is harder to estimate (Connor, Downing and Marston, 2017). However, one could reasonably anticipate increasing numbers of people with LLTCs living beyond childhood in countries with advancements in medical treatment, such as human immunodeficiency virus (HIV), and care similar to the UK.

Given its demographic infancy, limited empirical work has been carried out with this population as a whole (Abbott and Carpenter, 2016; 2014) and even less is known about the needs and expectations of young people with LLTCs on sex and relationships. Previous research in the area has tended to focus on *specific* conditions or disease types; see Frayman and Sawyer (2015), Kelly et al, (2017); Kelly, (2013); study on cystic fibrosis and Abbott et al's (2012) work on duchenne muscular dystrophy.

Much of the previous literature has focused on either the impact of the condition on sexual and reproductive health and/or the impact of sexual and reproductive health decisions on the health of the individual (Hordern and Street, 2007a; Hordern and Street 2007b). There is also an additional body of literature that addresses the issue of sexuality and disability in more general terms (for example, see Lund and Johnson, 2015; Liddiard, 2011; Earle and Church, 2004; Blackburn, 2002; Shakespeare, Gillespie-Sells and Davies, 1996). I argue that this research is timely and seeks to address a research gap. The objective of this research is to explore the experiences of young adults with different types of life-limiting or life-threatening conditions, addressing their intimate, sexual, relationship and reproductive opportunities, aspirations and expectations.

The purpose of this first, contextual chapter is to capture the transition points that are relevant and underpin this doctoral research. In using the term ‘transition’, I point to the theoretical framework that I have adopted throughout this research: a life course perspective, for example (Elder, 1985). I draw upon the related concepts in which this thesis is framed: transition theories (Van Gennep, [1909]1960), liminality, (Turner, 1969), biographical disruption (Bury, 1991; 1982) and the uncertainty about wellness, illness and end of life along the life course (Peou and Zinn, 2013). Please see Chapter 2. Through the lens of transition, I also explore my own doctoral journey, beginning with the rationale for my research, the questions I seek to ask, why, to whom, and the important documentary and governance arrangements required to achieve these goals. Importantly, transition includes the historical and philosophical changes that have occurred within contemporary policy and the political context over the last half century. This has seen successive changes in government and consequently transitions within their strategic policy imperatives and priorities.

The growth and development of palliative care services over the last six decades heralds another transition. Palliative care services – and particularly adult hospice services – emerged as an adult-based service (Clark 1998a; 1998b and 1998c) and have subsequently expanded to include the important needs of children and young people (Goldman, 2018; McNamara-Goodger and Feudtner, 2012). I suggest that we have reached another transition point. There are an emerging and growing number of young adults with LLTCs who have defied medical science and are now living far longer than ever anticipated (Norman and Fraser, 2014). Moreover, young adults with LLTCs are making an important transition from childhood to adulthood, and many are embracing important physical and emotional changes, often on an uncertain life course, and not least recognising their emerging sexuality (Together for Short Lives, 2015; Beresford and Stuttard, 2014).

The right to sexual experiences and fulfilment has been central to the politicising of sexuality for marginalised groups for over half a century, particularly in relation to

people with disabilities (Shakespeare, 2000). Talking about sex and relationships can be difficult, in so far as it is often considered a subject that is personal, private and taboo (The Open University with Together for Short Lives, 2016; ACT and Triangle, 2011). For young adults with LLTCs, discussing this subject may be more difficult because most of these individuals were not expected to live into adulthood. Yet their life course trajectory and expectancy may remain uncertain and limited (Norman and Fraser, 2014). They may be living with physical, sensory and/or cognitive impairments, as well as experiencing periods of acute and chronic illness and treatments (Abbott et al, 2016). Many young adults may have lived, or are still living, in residential settings and may have spent considerable time in hospitals or in hospices. On reaching adulthood, they may still be living with family, be reliant on carers and be less likely to live independently in comparison to non-disabled people of a similar age (Jepson, Abbott and Hastie, 2015; Together for Short Lives, 2015).

I shall argue throughout this thesis that the intersecting factors – sexuality, illness and an uncertain life course – imply a hypothesis of ‘cumulative disadvantage’ that is associated with a lack of both positional and participation opportunities (Levy and Bühlmann, 2016). In addition, there are implications for their emerging adulthood in relation to sexuality, intimacy, relationship opportunities and experiences. The lens of transition highlights the commonality and congruence through a number of disparate aspects of this research. These I use in this first contextual chapter to set the scene and then in subsequent chapters to explore the views, opportunities and experiences of young adults with LLTCs, and those who support them, as they relate to their relationship and intimate experiences. I place at the very heart of this research the lived experiences of sexuality in an emerging group of adults with LLTCs.

1.1 *Aims and rationale*

Care practitioners and the public are often reluctant to discuss sex and sexuality with young adults with life-limiting or life-threatening conditions, and particularly those near the end of life, or where a life course may be uncertain (Craig and Lidstone, 2012; Blackburn, 2002). This reluctance may be prompted by professional concerns around the sexual needs and expectations of people with LLTCs and the associated risks, including the potential for sexual exploitation by others (The Open University Sexuality Alliance with Together for Short Lives, 2016). There is often a corresponding reluctance on the part of young adults with LLTCs to share their experiences about sex and sexuality because of shyness or embarrassment, fear, a lack of privacy, or as a response to the reticence they perceive in others. This mutual avoidance of ‘talking about sex’ may result in a lack of expertise and understanding on both sides, particularly the specific issues related to the sexuality of people with LLTCs as they transition from childhood into adulthood (Liddiard, 2011; Blackburn, 2002).

Young adults with LLTCs have made it clear that they want sex and sexuality to be brought ‘out into the open’ and not ‘swept under the carpet’. Young adults want ‘to live life to the full’ for as long as possible (Watts, 2018); as advances in medical treatment, now enable them to live into adulthood.

This thesis aims to respond to these issues and I explore them from various, and sometimes competing, perspectives: on the one hand, young adults with LLTCs, and on the other, the partners, parents, and health and social care practitioners who support them. My starting point is to explore these subjects primarily with the young adults, seeking their views about sex, sexuality, relationships, intimacy and reproduction; what these mean to the individual participant, and how their views shape or are shaped by their own understandings, opportunities, personal experiences, and reproductive aspirations and choices. I aim to provide a forum for

participants with LLTCs to talk about sex and sexuality in a dignified and trustworthy manner, to be listened to, to be heard and not to be burdened by:

‘the idea that a young person’s life course is reduced through the presence of impairment.’ (Goodley, Liddiard and Runswick-Cole, 2017, p. 2)

From there, I pursue the other side of the conversation and explore the research questions with partners, parents and care practitioners, similarly seeking their views about sex, sexuality, relationships, intimacy and reproduction as they apply to, and are experienced by, the young adults with LLTCs.

This research respects young people’s choices and fundamental human rights, including sexual rights and sexual citizenship (Liddiard, 2018; 2011; De Than, 2015), implicit in Articles 2, 8 and 10 of the Human Rights Act, 1998, (see Appendix B). The aims, with a particular emphasis upon the views of young adults with LLTCs, are drawn together and articulated in the following research questions, which are explored in Chapters 4 to 7:

1. What are the views and experiences of young adults with life-limiting or life-threatening conditions (LLTCs) about relationships, intimacy, sex and reproduction?
2. To what extent are young adults, over 16, with LLTCs prepared through education and information, enabled to make relationships, sexual and reproductive choices?
3. What are the relationship opportunities and experiences for young adults with LLTCs?
4. What are the views of partners, parents, carers and care practitioners, who support people with LLTCs, on all of these subjects (Questions 1–3)?

These questions are intentionally broad and recognisably ambitious, and supported by prompts outlined in Appendix J.

1.2 *Research context*

This research is an evolving research journey, including a retrospective explanation of how and why the research developed and influenced the choices, fieldwork, analysis and my writing up. Progressive evolution and change is an inevitable feature of the doctoral journey (Barbour, 2014) and I argue that my research is no exception. Although the original research design and approach did not change significantly over time, some of the content, such as co-opting partners, at the request of two young adults, and addressing posthumous consent, led to amendments and discussions with The Open University Human Research Ethics Committee (HREC). These points are discussed in Chapters 3 and 8.

Throughout this thesis, I draw on health, social-scientific and legal literature, akin to my policy background. As stated in my introduction, I adopt a life course perspective which I believe suits the nature of this research and works well with my qualitative methodological approach. I have recorded, documented and transcribed verbatim 39 face-to-face interviews with 35 participants, at home or in a hospice (13 young adults, two partners, ten parents and ten care practitioners). I met some of the young adults with LLTCs on more than one occasion. Participants included young adults with cystic fibrosis (CF), duchenne muscular dystrophy (DMD), people with other neuromuscular degenerative conditions, certain cancers and genetic conditions, see Table 1 and Appendix I.

The young adult participants were predominantly, though not exclusively, white British, cisgender, mainly heterosexual, possibly asexual (discussed in Chapter 5) and

aged between 16 and 40 at the time of interviews, their average age being 26 years. Two young adults have died since interviews. All the young adults were identified as having either one or more LLTC, which were acquired either at birth, during childhood or adolescence. Some participants had marginal learning difficulties. Whilst recognising the importance of their needs, people with learning difficulties are not the focus of this particular research as other studies have been undertaken or are ongoing (Ledger et al, 2016; Abbott, 2016; 2015; Tilley et al, 2012a; 2012b).

At the time of interviews, all the young adult participants were living within the community, mainly with parents or carers at home, although many continued to attend 'short-break care', (see Appendix A), mainly provided by young adult hospices. My rationale for the selection of participants is detailed in Chapter 3, Section 3. This thesis is, among other things, a collection of the four groups of participants' evocative narratives. The interviews, set over a period in time, are located in England. The findings have been thematically analysed and shed light on an important and under-researched area of sexuality, specific to a growing number of young adults with different (but not all known) LLTCs who are now living into adulthood.

Why now?

There are several reasons as to why the sexuality of young adults with a range of LLTCs has not been specifically researched. First, as indicated in my introduction, until recently children and young people with LLTCs were not expected to reach adulthood (Abbott and Carpenter, 2014), so research about anything related to their adulthood, let alone sexuality, was not considered a priority (Craig and Lidstone, 2012). With advancements in medical science and technology, many young adults with various LLTCs are now surviving into adulthood, and some into their third or fourth decade, for example, men with duchenne muscular dystrophy (DMD) (Abbott, Jepson and Hastie, 2016), people with cystic fibrosis (Frayman and Sawyer, 2015), and those

surviving childhood cancer (Kelly and Vougioukalou 2017; Kelly, 2013). Second, given the young adults' anticipated greater life expectancy, they are showing a greater interest in sexuality, whilst recognising that their interest may impact on other individuals, particularly those caring for or supporting them (Jepson, Abbott and Hastie, 2015). Finally, this is difficult research 'to get right' and 'getting it wrong' could make things worse, importantly for the participants of the research, but also for the researcher. Sexuality is often regarded a 'taboo' research area, particularly for those on an uncertain life course (Taylor, 2012; Hordern and Street, 2007a, 2007b, 2007c, 2007d; Hordern and Currow, 2003) and can be a difficult discussion topic (Taylor, 2012; Liddiard, 2011).

1.3 *Personal transition: Why me?*

Both my academic and professional backgrounds have influenced the way that this exploration has been undertaken. These have underpinned the theoretical and methodological approaches I adopted. I came to academia as a very mature student, following a professional career. Thus, my background and interest were part of this thesis motivation. I have an enduring professional interest in the needs and experiences of under-researched groups, such as children and young adults with LLTCs.

The academic impetus and interest also arose from previous research at Charing Cross and Westminster Medical School, now Imperial College. This focused on the sexuality of young adults with two specific but interrelated life-limiting conditions: spina bifida and hydrocephalus (Blackburn, 2007, 2002; Morgan, Blackburn and Bax, 1995). Subsequently, as Director of Care Development for a national charity, I undertook an audit of 44 children's and young people's hospice services in the UK (Blackburn, 2010). Although sexuality was not its primary focus, that audit highlighted gaps in transition into adult life palliative care provision for people with LLTCs.

Over the last two decades, I have gained research experience under the broad heading of 'life course transitions'. Since 2012, the Association of Paediatric Palliative Medicine (APPM) and Together for Short Lives have collaborated to foster a research culture through a National Research Group, chaired by Professor Myra Bluebond-Langner, True Colours Professor in Children's and Young People's, University College, London. One strand of this work has been to establish a Doctoral Students' Taskforce, which I currently chair. The National group continues to assist research enquiry to ensure that children and young people with LLTCs and their families are consulted and are able to contribute to high-quality, policy-focused research. I remain an active member of this group and contribute to the academic and policy agenda related to transition into adult life, particularly the sexuality, consent and emotional wellbeing of young adults with LLTCs.

The inspiration behind this thesis also developed from other national and international policy work, spanning 25 years, witnessing the growing awareness and interest of people with LLTCs in sexual matters, and often their frustration at the lack of official acknowledgement and recognition of young people's sexual citizenship and agency (Liddiard, 2014b). During that period, I worked in clinical and policy roles within the public and third sector, as a Chief Executive and Policy Director. I mainly worked with young adults, over 16 years, with LLTCs who had complex health and social care needs. These roles kindled the desire to pursue a doctorate and thus add credibility to an area of research which is often viewed as taboo, and with suspicion and caution (Liddiard, 2018; Taylor, 2012; Liddiard, 2011). In 2007, a short secondment to the Adolescence Directorate, the World Health Organization (WHO), Geneva, enabled me to examine some service priorities for young people over 16 years with complex disabilities. WHO has undertaken considerable work to review the sexuality, human rights and legislation addressing the relationship and reproductive needs of both disabled and non-disabled people (WHO, 2015). Furthermore, having acquired cervical spondylitis myelopathy (CSM) in later life (see Appendix A), I not only

share the understanding and difficulties, but some of the neuromuscular degenerative features of certain LLTCs, albeit to a lesser degree.

Central to my approach is a desire to make an important contribution to research, leaving a legacy towards the end of my professional career that may assist international policy and practice. My research activity is predicated on this central aim. Throughout my career I have aspired to develop a reflexive, sympathetic, non-judgemental and professional approach when working with disabled people and interlocutors. I describe myself as a humanist researcher, who is respectful of gender, sexual orientation and human rights, and with a research emphasis:

‘that gives prime place to human beings, human meaning, and human actions in research. It usually also works with a strong ethical framework that both respects human beings and seeks to improve the state of human kind in a global context.’ (Sage Encyclopaedia of Qualitative Research, accessed 11th January 2018)

1.4 Transitions in determining terminological usage

The terminology employed in both clinical and social-scientific literature in relation to generic, palliative, end-of-life care, and specific life-limiting, life-threatening conditions, is sometimes confusing, complex and cumbersome and may distract from the important tenets of the research inquiry. This I seek to avoid. For ease of navigation, in this first chapter, I have set out to standardise terminology and to clarify some of the generic and specific terminology I use throughout this thesis and explain why, whilst detailing abbreviations and other generic and clinical terms in Appendix A. To adapt Einstein, I believe that terminology should be ‘as simple as possible, and no simpler’ (attributed to Albert Einstein, undated). Moreover, I try to exercise consistency and etiquette, avoiding perceived, subconscious pejorative language and

the dehumanisation of disabled people. Throughout, I refer to a *participant* or *person* or *adult* and seek to avoid, except in quotations, alternatives such as *patient*, *service-user* or *client*, as these seem to deny the uniqueness and individuality of the person with a LLTC by identifying them only in relation to their care-providers. These terms also underestimate the complex nature of individual needs and requirements beyond the LLTCs, particularly sexuality. For similar reasons, I prefer the phrases 'person who has *sustained* or *acquired* a LLTC' or 'person *with* a LLTC', rather than 'person who has *suffered* a LLTC'. 'Suffer' and its various permutations imply a punitive or passive conception of the LLTC and a problematised relationship between the LLTC, *suffering* and uncertainty. I refer to 'disabled person', 'disabled people' and 'people with disabilities', reflecting that disability is both part of, and a respected component of, a person's citizenship and sexual identity (Liddiard, 2011), as opposed to 'people with impairments' or 'handicapped people', which I regard as pejorative. Collectively, sometimes I refer to partners, parents, carers and care practitioners as 'supporters'. It is unavoidable that I use the terms 'death', 'dying' and 'end of life' as these relate to the theoretical life course perspective and uncertainty which I have adopted, and are relevant to the individuals as well as the data analysis in Chapters 4 to 7.

In the late 1980s, the terms 'life-limiting' and 'life-threatening' (or life-shortening) conditions replaced 'terminal' or 'dying children' (Eiser, 1993). Clinicians sought to describe an emerging and new population with distinct health and social needs (Stein, Wooley and Baum., 1989). The term 'life-limiting or life-threatening conditions' established a more conceptual framing of children's and young people's palliative care and to ensure that clinicians recognised and prioritised the needs of this population, their families and carers in a socially inclusive way (Goldman, 1994). These terms continue to be used today (Together for Short Lives, 2017). I suggest that the use of different terms to describe this particular population may continue to evolve as life-expectancy extends, treatments and technological interventions advance, and people live longer.

I argue that 'life-limiting' or 'life-threatening' are categories of the conditions (see Section 1.9 and Appendix A) and bring into question whether the name and knowing the type of LLTC alters or impacts on the young adult's illness experience, a feature I explore in Chapter 4. For the purpose of this research, I developed a specific definition for the young adult participants, see below. I was assisted by the National Research Group, see section 1.3. This is because the age range for this particular research is broad and there are different definitions of young adults used in the literature (see Appendix A) and Sawyer et al., 2018:

'People between 16 and 40 years who were born with or who acquired life-limiting or life-threatening condition(s) during childhood, adolescence or early adulthood; some of whom are living until their third or fourth decade but who are unlikely to reach old age.'

1.5 Transition and emerging adulthood

Over the last few years, empirical and theoretical understanding of early or the first stage of adulthood has undergone contemporary change (Tanner and Arnett, 2009). 'Emerging adults' during adolescence often process information differently from their older counterparts. Emerging adults may respond to more emotional situations and may be linked to the brain's maturity (Tanner and Arnett, 2009). I shall argue that the young adults in this research may emerge as adults at different chronological and physiological stages, and that this may be compounded by their LLTCs. Such 'transitions' can be complicated by deteriorating physical health and sensory and/or learning difficulties alongside normal developmental changes (Doug et al, 2011). I recognise that the legal age and policy definitions related to children, young people, young and older adults differ and these variations are outlined in Appendices A and B. Sawyer and her colleagues seek to avoid too narrow an interpretation of transition

from childhood to adulthood and include young adults in their mid-twenties, both with and without disabilities:

'The transition period from childhood to adulthood now occupies a greater portion of the life course than ever before at a time when unprecedented social forces, including marketing and digital media, are affecting health and wellbeing across these years. An expanded and more inclusive definition of adolescence is essential for developmentally appropriate framing of laws, social policies, and service systems [...]. Overly narrow age definitions miss the opportunities to be gained from implementation of the scope of cost-effective multisectoral interventions that are transformative for nations.' (Sawyer et al, 2018, p. 223)

Furthermore, the notion of a child or young person living a short life is deeply emotive (Goodley, Liddiard and Runswick-Cole, 2017). People are affected, often by the idea that a young adult's life course is reduced through the presence of a LLTC. The responses to young people with LLTCs are frequently shaped through societal ideas and practices linked to idealisations associated with quality of life, human productivity and, importantly, living life to the full. Society feels for the predicament of young adults with LLTCs and excuses them from the neoliberal imperatives of self-sufficiency, independence and sexual autonomy (Goodley, Liddiard and Runswick-Cole, 2017). Some participants regard the terms 'end of life', 'dying' and 'death' almost with dread, while:

'Cultural responses to these young people are shaped by dominant discourses associated with lives lived well and long.' (Goodley, Liddiard and Runswick-Cole 2017, p. 1)

Whilst Goodley and his colleagues here emphasise the desire ‘to live life to the full’ and not to be constantly reminded of impending or uncertain death.

1.6 *Sex(uality) as a shorthand*

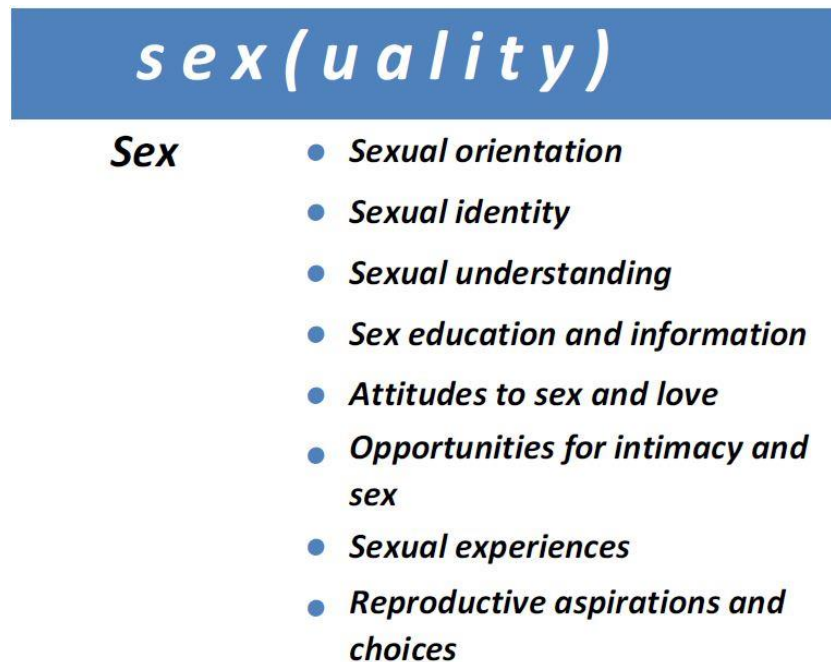


Figure 1: Sex(uality) used as a shorthand for the research.

In this research, I use ‘sex(uality)’ as a shorthand, to support the research question prompts, see Appendix J, to provide consistency, as well as avoid repetition in my interviews and analysis. It also serves as a model for my thinking, building upon, but different from, a sexuality acronym I previously used (Blackburn, 2002). Sex(uality) in this research includes the components outlined in Figure 1 and is used to discuss answers to the research questions I asked all four groups of participants (see Section 1.1). I restrict its use to discussion of the findings and separately refer to ‘sex’ or ‘sexuality’, and the different components outlined in Figure 1, when addressing other authors’ work. The components reflect some of the literature I explore in Chapter 2. I recognise that sex and sexuality have multiple interpretations (WHO, 2015) that

include both physical and non-physical aspects of sex(uality) (Liddiard, 2018, 2011). These differences and meanings are explored in Chapter 5.

1.7 *Transitions: The history and philosophy of palliative care*

There is a certain complexity in the literature around children's and adults' palliative care, see Appendix I, because there are similarities and differences in these services (Goldman, 2018; Clark, 1998a). There is now the emergence of a third group of young people with LLTCs who are on the threshold of adulthood. This third group is my primary focus in this thesis.

Over the last century, palliative care has developed as a unique and explicit specialism for all age groups, creating its own philosophy and often rendering it different from services provided in NHS health-care settings (Randall and Downie, 2010). The modern palliative care movement is still relatively new and developing. It largely earns its reputation from:

'the philanthropic, religious influences and approaches from early twentieth century health care practices which helped shape the twenty first century hospice movement.' (Clark, 2008a, p. 39)

Historically, palliative care has been devoted to providing expert, holistic care to people with LLTCs of all ages; care that serves the individual's goals for quality of life, not just quantity of life (Gawande, 2015). Previously, adult palliative care has primarily focused on end-of-life clinical care, particularly in adults (Clark, 1998a, 1998b). In the UK, its application is often seen as contentious and is often politicised in health-care debates (Gawande, 2015; Clark, 1998a, 1998c).

Both children's and adult palliative care services are premised on the holistic principles of physical, emotional and spiritual care, and the wellbeing of individuals and the families or individuals supporting that person. It is recognised that family members may also require care and support at various stages of caring for, or following the death of, a child, young or older adult (Hospice UK, 2013; Aldridge, 2008).

The various definitions and interpretations of palliative care are detailed in Appendix A, but in essence:

'The term "palliative" is derived from the Latin word pallium meaning a cloak. Palliative care aims to cloak the patient's symptoms and provide comfort even when treatments aimed at cure are no longer possible.' (Muckaden et al., 2011)

'The association with a cloak dates back to early Christian symbolism where the giving or sharing of a cloak was a first gesture of Christian care for those in need, St Martin (of Tours) sharing his cloak with the beggar being the most famous instance.' (See <http://www.stmartinschurchguernsey.org/about-us/the-story-of-st-martin/>, accessed 11 April 2018).

Adult palliative care services

There are differences as well as similarities in the philosophic underpinning of palliative care for children, young people and adults. Both adult and children's hospice care similarly embrace the philosophy and practice of palliation and holistic care for anyone with a LLTC, at any age or stage of life, who requires pain and symptom management (Beale, Baile and Aaron, 2005). Holistic care is a core principle of health and social care practice for all age groups and is recognised by professional bodies, such as the Nursing and Midwifery Council (2018). The philosophy of holism reflects the World Health Organization's (2001) bio-psycho-social approach to health and

social care, and its ethos requires care practitioners to assess these aspects in order to address the needs of individuals. If care practitioners are to use a holistic framework as the basis of caring and assessing needs, Taylor (2012) argues that sexuality should be an important feature within care plans. I return to this in Chapter 8.

Until recently, a notable difference between adult and children's palliative care services was that in adults, this was provided for a shorter period; often just the last few weeks of life. Its focus was on symptom control, primarily supporting older adults with cancer through the active withdrawal of treatment. Now older adults in the UK may continue to engage with palliative care services over a longer period and a wide range of conditions are included, in addition to cancer, including multiple life-limiting conditions, such as motor neurone disease, cardio-vascular conditions, multiple sclerosis and Parkinson's disease (Hospice UK, 2018). The nature of the LLTC may mean that young people and their families receive holistic care and support for many years, both in hospices and in community settings.

In the 1960s, Dame Cicely Saunders founded the modern hospice movement for adults. Her diverse training helped her to conceive and develop the model of holistic care at St Christopher's, London, in 1967, the first adult hospice in the world (Clark, 2008a). The modern hospice movement in the UK for both adults and children provides care for those who prefer to end their life at home, or in a hospice, but not in hospital (Clark, 2008a). Hospices also support people's families, especially with bereavement support. Most hospice care (80%) in the UK is provided in community-based settings, such as home care/hospice-at-home, outpatient services and hospice day-care (Hospice UK, 2017). The remainder is mainly in hospital or other residential settings. In a recent review of UK hospice services for both adults and children, the Care Quality Commission rated hospices overall as providing an extremely high standard of care, arranging good quality services with attention to dignity and respect, and a caring and positive approach for the individuals and families using them (CQC, 2014).

Children's palliative care services

The number of children and young people who die from a LLTC is relatively small compared with the number of adults. More than 5,000 children (aged up to 18 years) die every year in England and Wales, about half of whom die from LLTCs (Fraser et al, 2013; 2012). Children's palliative care, although sharing some similarities, has distinct differences from adult palliative care (Aldridge et al, 2017; Goldman, 1994). The main difference is that, compared to older adults, children with LLTCs usually access palliative care services, either in hospital or via a hospice or community service, over a longer period of time (Goldman, 2018).

In 1982, Sister Frances Dominica, a nurse and Anglican nun, created Helen House in Oxford, England, the world's first free-standing hospice for children requiring holistic palliative care. The Helen House philosophy focussed on providing short care breaks, as well as end-of-life care for children and bereavement support for families (Burne, Dominica and Baum, 1984). Seven years later, she also founded Douglas House, the first young adult hospice, in response to increasing numbers of young people with LLTCs surviving into adulthood and which has now closed.

In 1985, Dr Ann Goldman, a paediatrician at Great Ormond Street Hospital, developed the first paediatric palliative care programme in hospital to support children with malignant diseases, and their families. She and two clinical nurse specialists cared for children throughout the disease trajectory, from diagnosis to death, to manage symptoms and to work with local care providers to facilitate both home and hospital care as required (Goldman, 2018). Goldman recently reflected that changes occurring both in society and medicine during the 1980s have widely contributed to the development of children's palliative care over the last few decades (Goldman, 2018). Paediatric palliative care has evolved to become a distinct specialty that involves multidisciplinary teams working across numerous organisations, including hospitals,

community settings and children's hospices (Aldridge et al, 2017; McNamara-Goodger and Feudtner, 2012).

Holistic, palliative or specialist palliative care, see Appendix A, is required by children and young people with a wide variety of LLTCs, including cancer, organ failure, metabolic, genetic or degenerative illnesses, or static conditions such as severe cerebral palsy or epilepsy, where life-threatening complications may mean that survival into adulthood is less likely, but, for others, such as people with cystic fibrosis, specialist palliative care may be needed for many years (Aldridge, 2017).

This encompasses symptom control for the young person, and emotional and psychological support for the child and family. It also addresses their practical, financial and spiritual needs from the time of diagnosis of a LLTC throughout the child's life, and extending to bereavement support for surviving family members after the child or young person has died. Necessarily, this means providing *individualised* care (Aldridge et al, 2017).

Over the last twenty years, similar models of palliative care have developed internationally (Marston and Chambers, 2012). The UK continues to provide international leadership in addressing palliative care services for children, young people and older adults (Goldman, Hain and Liben, 2012).

Every child or young person's medical, social and psychological circumstances are unique, and each and every narrative that unfolds may be particularly distressing for the young people and families who support them.

1.8 *Transition: The emergence of palliative care for young adults*

Children's and young people's palliative care is not just for those who are about to die. It is person- and family-focused and needs-based (McNamara-Goodger and Feudtner, 2012). Children and young people, diagnosed with neurological and/or degenerative conditions, such as Duchenne Muscular Dystrophy (DMD), may receive or access specialist palliative care from birth and increasingly into their third or fourth decade, bringing into question whether palliative care for young people with some LLTCs should be referred to as long- term or complex care provision.

Until recently, the UK palliative and hospice care provision recognised two distinct population groups: children and young people, and (older) adults (Together for Short Lives, 2015). As explained in Section 1.6, both populations require similar and different types of services and both groups of people may present with an uncertain life course trajectory.

With advances in medical science and technology, this picture is becoming more complex. Young adults with LLTCs are a distinct and emerging population with particular needs that differ from children (Fraser et al, 2014; Beresford, 2004).

At the time of writing, there are around 300 adult hospices in the UK (Hospice UK, 2017), and 53 children's and young people's hospices. Children's hospices support babies, children and young people with a wide range of complex LLTCs, including neuromuscular degenerative conditions, genetic disorders and cancers.

The duration of engagement with hospice services is lengthening both for children and adult services and these provisions are evolving to cope with these demands. However, there is a gap in provision for young adults, aged 16 and above, who are no longer children but distinct from older adults. The demand for care among this group

is far greater than the supply, possibly a side-effect of the increasing survival of young people with LLTCs. There are currently six hospices providing dedicated day, residential or hospice-at-home care for young adults over 16 years with LLTCs in the UK (Together for Short Lives, 2017).

Children's and young adults' hospices usually have an upper age limit for those for whom they provide services, as recommended by the Care Quality Commission. Devanney and Bradley, (2012) reported that around 8,000 families utilised either a children's or young adults' hospice service. This represents a relatively small proportion of the reported 49,000 children and young adults with LLTCs in England (Fraser et al, 2013; 2012).

Young adults who might previously have used a children's and young person's hospice or have been supported by family at home may later require direct entry to an adult hospice or residential service (Aldridge et al, 2017). Children's hospices frequently provide residential weekends where young people and young adults not only receive treatments and residential 'short breaks', but the opportunity to socialise with their peers. The children's and young adults' hospices also provide considerable support for siblings, offering residential respite or short breaks in hospices for both the young person with a LLTC and their families.

This distinct third group, the emerging young adults with LLTCs who are transitioning from adolescence to adulthood, are seeking independence; increasingly, they want to socialise, and talk about and experience sex(uality), all in the shadow of their ongoing LLTCs (Abbott, Jepson and Hastie, 2016; Abbott and Carpenter, 2014) but on an uncertain life course. This presents a new dimension to any holistic service requirement, as yet unprovided-for, where adolescents fall somewhere 'betwixt and between' (Turner, 1969) adults and children's services.

1.9 Classification and aetiology of LLTCs

Table 1 provides the four main categories of life-limiting and life-threatening conditions that are described and widely adopted internationally by both researchers and practitioners. It provides a brief aetiology of LLTCs and specific conditions affecting both males and females.

Category of LLTC	Definition
Category 1	Life-threatening conditions for which curative treatment may be feasible but can fail. Access to palliative care services may be needed when treatment fails or during an acute crisis, irrespective of the duration of that threat to life. On reaching long term remission or following successful curative treatment, there is no longer a need for palliative care services. Examples: cancer, irreversible organ failures of heart, liver, kidney.
Category 2	Conditions where premature death is inevitable. There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: Cystic fibrosis, Duchenne muscular dystrophy.
Category 3	Progressive conditions without curative treatment options, where treatment is exclusively palliative and commonly extend over many years. Examples: Batten's disease, Mucopolysaccharidosis.
Category 4	Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health complications and likelihood of premature death. Examples: severe cerebral palsy, multiple disabilities such as those following brain or spinal cord injury, complex disability with high levels of health care needs and high risk of unpredictable life-threatening episodes.

Table 1: The four main categories of life-limiting and life-threatening conditions. Reproduced with the prior permission of *Together for Short Lives* (2018).

The life-limiting or life-threatening conditions relevant to the young adult participants in this research are outlined in Appendix I. People with life-limiting and/or life-threatening conditions are often addressed together but the research highlights a

distinction between the two groups. Policy rhetoric is distinct from the reality of what people experience in relationships (Abbott, Carpenter and Hastie, 2015).

Given the increasing numbers of children surviving into adulthood with a LLTC, transition to adult services for the emerging adult with palliative care needs is a contemporary concern. Such 'transitions' can be complicated by deteriorating physical health and sensory and/or learning difficulties alongside normal developmental changes (Doug et al., 2011). Nationally, published data for young people aged 19 to 40 years with palliative care conditions is patchy (Fraser et al., 2014; Hughes-Hallett, Craft and Davies, 2011). Some charity registries record the numbers of people with specific conditions, such as the Cystic Fibrosis Trust and the Muscular Dystrophy Campaign. This is not the case in every charity. Individuals or families must now offer their agreement to register the specific condition and share any data (Harris, 2018). Currently not all charities have access to precise data about the people they support and the implication of Data Repositories and data sharing is discussed further in Chapters 3 and 8. Given the ambiguity and consensus around terminology discussed in Section 1.4, it may not be surprising that until now there has been no central register of people with LLTCs in the UK (Hain et al., 2013a; 2013b; Savage, 2011; Craft and Killen, 2007).

1.10 Structure of this thesis

In this contextual first chapter, I have introduced my research, the rationale, its significance and my personal reasons and desires for undertaking it. I have introduced my theoretical and methodological approaches, and presented an historical overview of adults', children's and young people's services and some of the challenges around terminology. I have considered the potential gap in addressing the specialist palliative care requirements of emerging adults with LLTCs.

In Chapter 2, I review the literature in two main sections. I review the meanings of sex and sexuality, and the international empirical, legal, policy and grey literature related to intimacy, sex and sexuality. I focus on disabled people and particularly those with life-limiting or life-threatening conditions. I explain my rationale for drawing on contemporary theoretical resources to enrich the themes of uncertainty, transition theory, liminality and biographical disruption.

In Chapter 3, I outline the methodological approach and methods I have used and their suitability for this thesis. I also provide background to the participants of the study.

Chapter 4 introduces the four data chapters, apprising the reader of conditions, clinical care, and the stories around how the participants- young adults with LLTCs- were first diagnosed and how this impacts on the research questions; and their lives, including their sexual lives. This is more than a scene-setting chapter. The LLTCs and their impact on individuals are defining features of the research and merit separate discussion. I discuss why the findings related to uncertainty, liminality, transitions and biographical disruption along the life course, provide the springboard to explore the research questions, addressed in Section 1.1, and then pursue them in greater depth in the following three data chapters.

In Chapter 5, I explore the distinction between ‘formal’ and ‘informal’ sex and relationship education (SRE), the participants’ preferences, and what this means for the sexual citizenship of the research participants. I consider whether the presence or absence of either ‘formal’ or ‘informal’ SRE influences or impacts on how young adults with LLTCs are able to engage in social, intimate and sexual relationships.

In Chapter 6, I discuss the meanings, opportunities and experiences of sex(uality) with all groups of participants but focusing on the young adults’. I explore whether the

presence of a LLTC condition creates or is a barrier to relationship opportunities and/or experiences.

In Chapter 7, I explore the specific issues and experiences of the young adults with LLTCs in relation to their reproductive aspirations, loss and the future within an uncertain life course. I address the findings related to the uncertainties about the impact of the LLTC during puberty, and emerging adulthood, including how all four groups of participants obtain information related to genetics – through their own research, from practitioners and genetic counsellors, or via the internet or other sources.

In Chapter 8, I recapitulate the key findings that have emerged from both the empirical and methodological data and provide some final reflection on the PhD journey, before drawing together the final conclusions and implications for policy and practice.

1.11 Conclusion

Within this first chapter I have laid the foundations and parameters for this thesis. I have introduced why this research is now important, and why young adults with LLTCs as a population are a growing and under-researched community in relation to their sexuality.

At the heart of this thesis are the young adults with LLTCs. Throughout the thesis, I shall strive to explore approaches and strategies that are both inclusive and serve a social justice and human rights agenda, giving a voice to those who are sometimes disenfranchised within society (Liddiard, 2018; 2011). I shall also focus on the intersectionality and lived experiences of sex, sexuality and disability within an

uncertain life course, blending intellectual originality, innovation and rigour (Dean et al, 2017) with the literature review which follows next.

Chapter 2 Literature review

Introduction

This chapter summarises the key arguments as to why the sexuality of young adults with LLTCs is becoming an increasingly important feature in the lives of this group, albeit that their life course is uncertain. The chapter is divided into two halves, drawing on two bodies of international literature in order to make clear the terrain to which my research and its findings may contribute. The first half of the chapter, up to Section 2.3, is divided into five sub-sections and focuses on the empirical literature related to the sexuality of disabled people and adults with LLTCs. First, I introduce the contemporary meanings of sex and sexuality, sexual agency, intimacy and citizenship as they apply to people with LLTCs in the context of this research; second, I explore the emerging adult with a LLTC in the context of adolescence; third, I look at the empirical literature related to the sexuality of people with either life- limiting or life-threatening conditions; fourth, I explain the legal literature related to the sexual rights and ‘protection from harm’ considerations, and finally, I consider the literature related to reproduction, reproductive choices and loss relevant to people with LLTCs (Earle, 2014; Earle, Komaromy and Layne, 2012; Earle and Letherby, 2007).

In the second half of the chapter, I outline a body of literature related to the life course perspective, drawing primarily on the work of Elder (2000, 1998, 1995, 1978) and the related theoretical concepts in which this thesis is framed: transition theories (Van Gennep, [1909]1960), liminality (Turner, 1974, 1969), biographical disruption (Bury, 1997, 1991, 1982) and uncertainty (Peou and Zinn, 2013; Penrod, 2007; Earle 2014 and Charmaz, 1999). I also explore the intersectionality between areas often regarded as sensitive and taboo: dying, end of life, sexuality and disability (Earle and Blackburn, under review). Finally, I bring together the literature to explore how a life course perspective provides an insightful structuration into the sexual, intimate and

lived experiences of young adults with LLTCs through different viewpoints and lenses, and frames the specific areas of this research inquiry.

In each section, I use these arguments to examine the ways in which the sexuality of a growing number of adults with LLTCs requires both acknowledgement and prioritisation in the policy, practice, clinical and the UK's sex education curriculum (SRA), discussed in Chapter 8. I have adopted a multidisciplinary approach to this review. I draw on health care and clinical literature, such as Kelly (2013) Taylor (2012), and Hordern and Street (2007a); social scientific literature (Aldridge et al, 2017), legal literature (De Than, 2015), as well as UK statutory provisions, see Appendix B. Drawing on a life course perspective (Elder, 2000, 1995, 1978), I refer to medical sociology (Denny, Earle and Hewison, 2016; Beresford and Stuttard, 2014; Earle, Komaromy and Layne, 2012; Beresford, 2004), policy, and grey literature to inform my arguments (Together for Short Lives, 2015; Watts, 2018; Marie Curie, 2012).

This literature review is the knowledge base which my thesis aims to build upon. It represents an account of the evidence and wisdom currently available in my field of research. Drawing on a multidisciplinary approach presents its own challenges, but I have found it helpful in explaining phenomena and the exploration of experiences from the four different groups of participants in this research (the young adults, partners, parents and care practitioners). I have outlined my operational literature review, design and strategy in Appendix C which resulted in over 1300 searches and references incorporated into my ENDNOTE library.

2.1 *Sexuality, sexual agency and intimate citizenship*

Sexuality

There are several conceptual interpretations of sex and sexuality within the academic literature, which include reference to, but do not solely focus on, sexual behaviour (Liddiard, 2018, 2011; Taylor, 2012, 2011). Sexuality does not exclusively feature the biological and physiological components of sexual behaviour and reproduction (Hodge, 1995). Human sex(uality) includes both sociological and psychological aspects as to how people express and experience themselves as sexual individuals (The Open University Sexuality Alliance, 2016). For this thesis, I have adopted the World Health Organization's (WHO, 2015) framing of sexuality as I believe that its breadth relates to the diversity of the four groups of research participants. It also acknowledges sexuality as being central to human existence and its relationship between sexual health, holistic care and reproduction (WHO, 2015). Please see, Appendices A and B for the various definitions of sex(uality) and intimacy.

Although WHO, 2015 recognises that sexuality should be included as part of holistic palliative care, Taylor (2012) argued in her thesis related to adults with motor neurone disease and cancer, that there is insufficient research evidence to usefully support health care professionals to address the sexuality of adults with LLTCs. Taylor's findings highlight that the experiences and concerns of people diagnosed with a life-threatening illness, such as cancer or motor neurone disease are recognised but that issues related to sexuality at the end of life are not always addressed (Taylor, 2012). The End-of-Life Strategy (Department of Health, 2008) advocated identifying individuals' wishes and preferences regarding their care, but did not include how care practitioners should support people to experience human sexuality 'and express themselves as sexual individuals' (The Open University with Together for Short Lives, 2016, p, 8).

Sexual Agency

There are several scholarly interpretations of sexual agency (Liddiard, 2018, 2011; Gill, 2008). In essence, 'sexual agency' is the freedom and choice to make sexual decisions, without coercion. Kimmel (2018) argues that 'when discussing sexual agency, you have the ability to define yourself sexually, the ability to choose whether or not you want to experience sexual activity, the ability to choose how you want to engage in sexual activity, and the ability to stop or refusing any sexual activity' (Kimmel, 2018, online).

However, Liddiard (2011) notes that disabled people are often considered to lack sexual agency in their lives and are therefore often denied such choices. Furthermore, both sexuality and sexual agency are predominantly constructed through a non-disabled, hetero-normative 'ableist' lens and sexual choices are often ignored (Kelly and Vougioukalou, 2017; Liddiard, 2011). The sexual agency of disabled people has largely emerged from political debates over the last few decades around the social model of disability (Shakespeare, 1996) where the sexual lives of both disabled men and women were not regarded as important or prioritised and that sexuality is viewed as phallogentric, reinforcing the centrality of physical performance (Shakespeare, 2000).

There is an increasing literature suggesting that 'asexuality can be a benign and immutable sexual orientation rather than a pathology' (Lund and Johnson, 2015). Along with a growing movement for recognition among self-identified asexual people, individuals with disabilities have long been working to have their sexuality recognised and given legitimacy (Przybylo, 2013). People with disabilities, including those with LLTCs, have been wrongly assumed to lack sexual desire and function. Consequently, those involved in the disability/ sexuality movement are opposed to the idea of asexuality in the context of disability, seeing it as an inappropriate label which has

been oppressively imposed on disabled people by society (Milligan and Neufelt, 2001). Within a 'normative' life course, it is hoped that young people will exercise agency to make personal choices throughout their lives and this should include choices about sexuality. This study has indicated that this was not always the case for people with LLTCs.

Sex has been described as multifaceted, involving more than just the biological and physiological components of sexual behaviour and reproduction (Taylor, 2012). Sex includes both psychological and sociological aspects of how individuals relate to themselves and the world surrounding them (Hodge, 1995). Consequently, the terms 'sex' and asexuality may be problematic.

Richardson (2017) advocates the inclusion of disabled people, and lesbian, gay, bisexual, trans or queer preferences (LGBTQ+) into the sexual agency debate. Furthermore, there is not only a growing awareness of the individual's sexual rights, but recognition that people also have the right to choose to be asexual.

'Asexuality can be a benign and immutable sexual orientation rather than a pathology.' (Lund and Johnson., 2015, p. 126)

Whereas Liddiard (2011), like Brown (1995), argues that disabled people are erroneously regarded as:

'Asexual, oversexed innocents, or perverts' (Brown, 1995, cited in Liddiard, 2011, p. 125).

Increasingly, self-identified asexual people and particularly those with disabilities are seeking to be recognised and legitimised (Pryzbylo, 2013). Yet there are those who view asexuality with suspicion. Until recently, society assumed that disabled people

were asexual and that this assumption was indiscriminate, inappropriate and oppressive (Liddiard, 2018, 2011). Disabled people were often perceived as asexual, often infantilised and in need of protection from harm or abuse (Liddiard, 2018; Milligan and Neudfelt, 2001). In contrast, Gill (2008) argues that agency is based on the assumption that humans are not passive recipients of a pre-determined life course but can make decisions that influence the shape of their lives. Decisions may be influenced by individual orientations to the situation, with some decisions requiring more intense focus on the present, and others influenced by long-term goals that may or may not include sexual agency.

Intimate and Sexual Citizenship

Linked to sexual agency are the important concepts of sexual or intimate citizenship. From a sociological perspective, discussions about sexual citizenship may lead to different constructions of citizenship, including the recognition of both public and private practices (Richardson, 2017; Weeks, 1998). While there remains no singular definition of sexual citizenship (Richardson, 2000), a common understanding is that sexual citizenship refers to the claims to (sexual) rights that are made by a sexual minority group (Richardson, 1998; Weeks, 1998). Other understandings are that sexual citizenship is 'about enfranchisement, about inclusion, about belonging, about equity and justice, about rights balanced by new responsibilities', Weeks, 1998 cited in Bamforth, 2012, while Plummer (2003) utilises the notion of intimate citizenship (rather than sexual citizenship) because it centres claims to rights of public and private intimacies which extend beyond the erotic and the sexual. These different constructions highlight the norms of sexual citizenship, and indicate the importance of understanding the liberal concepts of sexual citizenship (Richardson, 2017; Bamforth, 2012).

Richardson (2017) argues that sexual citizenship is multifaceted and subject to different interpretations. While Richardson acknowledges that there is still no singular definition of 'sexual citizenship' (Richardson 2017; 2007; 2005; 2004; 1998), it is regarded as the sexual rights of a minority group (Liddiard, 2011; Richardson, 2017; Richardson, 1998). It may include 'social inclusion, enfranchisement, equity and justice, belonging and responsibility' (Weeks, 1998, p.39) and It is also about what Weeks describes as:

'the tangible but forceful reality of social existence and of all social relations.'
(Weeks, 1986, p. 7),

Over the last few decades, sexuality has emerged in debates about citizenship. Richardson (2017; 2007; 2005; 2004; 1998), Plummer (2003), Weeks (1998), and Evans (1993) have advanced such debates. Until recently, sexual citizenship mainly focused on the 'normative' assumptions about sexuality (Richardson, 2015; 1998; Canaday, 2011; Bell and Binnie, 2000; Weeks, 1998), as well as 'hegemonic, married, heterosexual practices' (Richardson, 2017, p. 204) with little reference to LGBTQ+ inclusion and issues pertinent to disabled people. Richardson, 2015, argues that this is due to the fact that:

'the operations of power and the role of social institutions that sustain gendered and sexualised inequalities are disguised, which makes addressing them [gender and LGBT] more difficult to discuss'. (Richardson, 2017, p. 214)

Richardson (2007) argues that gender and sexuality need to be examined together, with gender taking precedence over sexuality and the acknowledgement of LGBTQ+ in sexual citizenship within this debate (Richardson, 2017; 2004) as well as the importance of their legal and sociological constructs.

Liddiard (2016), Plummer (2003; 1995) prefer to use the term 'intimate citizenship' in preference to 'sexual citizenship':

"Intimate citizenship" [is] a young person's rights to be able to access intimacy and "the control [or not] over one's body, feelings, relationships (Plummer, 2003, p. 14)

Plummer (2003) argues that intimate citizenship relates to both public and private intimacies, extending beyond 'normative' sexual and erotic assumptions. Liddiard (2015) similarly recognises that 'intimate citizenship' is the right for a disabled person to experience intimacy, but that this is frequently ignored by those working with disabled people on the assumption that disabled people are not capable of having a sexual relationship or wish to experience sexuality and desire. These perceptions are also heightened by societal attitudes towards the sexual expression of disabled people and in which Liddiard particularly notes that disabled people are wrongfully:

'presumed to lack the capabilities and capacities to embody as well as the agency to love and be loved by others and build their own families, if they so choose.' (Liddiard, 2011, p. 215)

It is noteworthy that WHO (2015) does not specifically refer to sexual, intimate citizenship or intimate rights in its report on Sexuality, Human Rights and the Law but acknowledges the complexity of the intersectionality between sexual health, human rights and the law are complex requiring further discussion (WHO, 2015).

In summary, whilst the recognition of sexual citizenship, particularly by Richardson and her contemporaries, have foregrounded matters that have previously been ignored, such as accounts of citizenship, reproduction and the recognition of the relationship rights of disabled people and LGBT communities (Richardson, 2017;

Sabsay, 2013), the literature still leaves unquestioned gaps related to the liberal western frameworks of citizenship, particularly in marginalised groups (Richardson, 2015) such as people with LLTCs. Individual choice is an important feature of western, neoliberal citizenship which manifests in sexual and intimate citizenship. This focuses on the:

‘right to choose – your partner; whether to marry or not; to have a child or not; your sexual activities.’ (Richardson, 2017, p. 216)

and recognises that:

‘people who often have little control over their bodies, feelings, relationships; ... and few socially grounded choices about identities, gender experiences, erotic experiences.’ (Plummer, 2005, p. 93)

In this section, I have sought to introduce critiques that underpin where sexual or intimate citizenship fit today, stemming from a predominantly heteronormative literature, but now including LGBT+ and disabled communities (Richardson, 2015). I suggest that the concept of sexual citizenship has traction and validity within this thesis in addressing the sexual rights of people with LLTCs.

2.2 *Emerging adulthood*

Emerging adulthood is a distinct and critical life stage in young people’s lives in the development between adolescence and young adulthood. This is extended over time, typically from ages 18–25 years (Tanner and Arnett, 2009). It has been described as a key period when significant life events are likely to occur (Grob, Krings and Bangerter,

2001). It differs from adolescence and the period that follows it, and is historically embedded and culturally constructed (Arnett, 2000). Emerging adulthood focuses on the personal and psychological experiences, feeling in-between, uncertainty, instability and the development of an identity (Tanner and Arnett, 2009), and relates to the liminal and uncertain aspects I explore in Section 2.4.

Whilst Arnett (2000) recognises the important role of society in understanding emerging adulthood, Abbott and Carpenter (2016, 2014 and 2012) note that there are significant challenges associated with planning for and living life as an 'unanticipated adult' with a LLTC. These not only include living with both the physical and emotional aspects of a LLTC, but being able to talk about friendships, sex and relationships. Because young people are now emerging as unexpected survivors (Abbott and Carpenter, 2016), I suggest such matters have not often been explored with this group, for fear of upsetting family members as well as the taboos often associated with both sexuality and end-of-life discussions.

Beresford (2004) acknowledges the highly problematic and challenging concern of the transition of young disabled people from children's services to adult services, and from childhood to adulthood. For both able-bodied and people with LLTCs, moving from childhood to adulthood may be a time of severe upheaval, instability and disruption. At a time of increasing autonomy for able-bodied teenagers, the experiences of people with LLTCs emerging into adulthood may be in sharp contrast. Philp and Duckworth (1982) note that some disabled teenagers, whilst seeking to achieve normality, are constrained by mobility and access difficulties, often alien to their non-disabled peers.

Adolescence is 'a rite of passage' that is both a biological reality and a period of time that has particular significance in different sociological contexts (Tanner and Arnett, 2009). Adolescence usually lays the foundation for subsequent learning and

development. It is usually a period of rapid physical development, brain growth and emotional development for heteronormative people, as well as social exploration for most non-disabled people. These changes may include physical, psychological, spiritual and sexual desire but such experiences may differ for young people with different LLTCs (Amery, 2016).

Adolescence is also a time when decision-making is particularly heightened by emotions and social factors rather than practical reasoning (Day, 2016; Blakemore and Robbins, 2012), and where often non-disabled people are influenced by their peers to engage in risk behaviours. The influences of parents in 'important decision-making about young people's serious illnesses' may also heighten emotions and tensions (Day, 2016), particularly during puberty.

Adolescence is often a time of mood swings, intense feelings, higher risk-taking, lower impulse control, broad role exploration and an increased sense of vulnerability for heteronormative young people. All of these affect how adolescents experience transition from childhood to adulthood.

For those with LLTCs, hopes for a normal and healthy adolescence may be changed or interrupted because of their condition(s), which impact on many aspects of their lives, such as self-image and self-esteem (Thomas, Bax and Smyth, 1989), as well as uncertainty about the future. Kelly (2013) noted that for young people following cancer treatment, their transition to adulthood may be disparate in terms of physical, emotional, and social needs. The rate at which sexual maturity is reached may not follow a definite course, highlighting the importance of personal and individual assessment in cancer rehabilitation programmes for adolescents.

Precocious puberty

For some young disabled people, puberty may occur earlier; precocious puberty or later than non-disabled people, (see Appendix A). This may be before age seven or eight in girls, and age nine in boys. In girls, this may include any of the following before seven or eight years: breast development, pubic or underarm hair development, a rapid growth in height, start of menstruation, acne and a mature body odour. In boys, signs before nine years of age include: enlargement of the penis or testicles, pubic, underarm or facial hair development, rapid growth in height, deepening voice, acne and a mature body odour (The Open University Sexuality Alliance with Together for Short Lives, 2016).

Frayman and Sawyer (2015) report that a hundred years ago that G. Stanley Hall, the father of adolescent health, proposed that the period of adolescence should be extended to 24 years of age because of the extent of growth that occurs over this period affirm this rationale. Some young people with LLTCs are less likely to achieve independence. This may be because of the pressure and demands of their medical care and treatments, the physical limitations imposed by their condition and the ongoing parental or care involvement. This may appear to stifle a young person's independence (Craig and Lidstone, 2012; Beresford, 2004; Blackburn, 2002).

Socialisation, experimentation and development of social skills are necessary features of adolescent growth, development and sexual identity. Self-esteem, body image and development of sexual ego and moral identity are crucial to a child's smooth transition to adulthood (Blackburn, 2002). There have been a number of studies which imply that disabled children and adults are impoverished in social experience and in interpersonal relationships (Dorner, 1990; Anderson and Clark, 1982). Impoverishment not only leads to difficulty in the establishment of intimate relationships, but also curtails the development of social skills (Thomas, Bax and Smyth, 1989).

They argued that a sense of identity and feelings of autonomy are two key developments during adolescence and it is through social interaction that the making and breaking of many relationships occur (Thomas, Bax and Smyth, 1989). They suggest that the sense of identity changes in response to differences between how people perceive themselves and how others perceive them. For non-disabled people, the social activities of the young adult increase quite substantially in the early teens; peer group activities will predominate, usually, but not always, with the same sexual orientation, followed by more one-to-one relationships, which tend to become deeper and more involved. It is through social activity and making friends that the young person is able to confirm their personality, identity and sexuality. Experimentation within this context allows for growth and maturation. During this time, an adolescent's reactions are vulnerable to the responses of those in their environment, who may include teachers, parents, carers and his/her peer group (Anderson and Clark, 1982). It may be difficult for a non-disabled adolescent to achieve a satisfactory social and sex life; they have to overcome parental authority and disapproval, financial restrictions, and educational demands before finding a compatible person or group (Beecham, 2008; Blackburn, 2002). For the disabled teenager, such issues are less important than the difficulties caused by restricted mobility, illness and personal acceptability. The likelihood of the disabled adolescent facing increased isolation at a time when their social circle should be broadening has been well documented (Liddiard, 2018; Day, 2016; Beresford and Stuttard, 2014; Beresford, 2004; Blackburn, 2002).

This may leave the young disabled person in a state of unknowingness as to what extent, if at all, they will be able to function as ordinary sexual and social beings. Physical disability, in addition to chronic illness and social stigma, may cause social and sexual isolation. The everyday difficulties faced by the non-disabled adult are further compounded for the disabled person, for whom self-identity and the achievement of independence (within the parameters of a LLTC) are important (Blackburn, 2002).

The degree of self-satisfaction, personal fulfilment and stability any young person experiences upon reaching full maturity in adulthood is frequently determined as a measure of how that individual has learnt to adapt to, cope with and manage the experiences of their earlier years. However, it should also be said that the measure of a young person's self-esteem is also determined in a more dynamic way; that is, by feedback from peer groups and family, and the level of social acceptance and integration, together with the amount of independence and autonomy allowed both within the family structure and society (Blackburn, 2002).

Self-concept and identity are particularly important aspects of social development. Adolescence is a period when both non-disabled and disabled people are particularly sensitive about appearance and how others think of them (Neinstein, 2016). Undoubtedly, physical and communication difficulties lead to social disability. Dorner's (1990) study of adolescents with spina bifida indicated a high rate of depression. He noted that depressive feelings were exacerbated for both male and female groups by poor mobility, the severity of the disability and social isolation. Similarly, Beresford, Harrison and Wilson (2002) noted an increased rate of depression among adolescents with complex disabilities. The authors attributed this to lack of independence from the family and a lack of peer group interaction because of the young person's disability.

Several studies support the need for follow-up counselling and support to help address the vulnerability and the long-term implications following treatments for cancer and other LLTCs, in relation to the individual's self-esteem, body image, sexuality, confidence and social relationships (Kelly, 2013; Blackburn, 2002).

2.3 *Empirical research: sexuality and disability*

As stated in Chapter 1, young adults with LLTCs are living longer largely due to advances in medical technology (Fraser and Norman, 2017; Beresford and Stuttard,

2014). While research has begun to focus on the broader social needs of this group of young adults, it has largely focused on other aspects of personhood, including gender (Abbott et al, 2016) but has not always addressed their sexuality. Historically, disabled people have been constructed as genderless, with gender presented as a troubling and troublesome variable (Liddiard, 2011; Shakespeare, 1996).

Sexuality and people with life-limiting conditions

There have been studies conducted on the sexuality of people with acquired physical disabilities, particularly spinal cord injuries but a paucity of research specifically addressing the sexuality of young adults with a variety of LLTCs (Hodge, 2005). It is only recently that the sexual needs and rights of people with congenital physical disabilities have been acknowledged (Taylor, 2012). She suggests that a concern for people with physical disabilities sometimes relates to parental desires for them to lead a 'normal' life, contrasted by a parental need to 'protect' the young person with a LLTC from physical and emotional harm.

There is a growing literature about the sexuality of disabled adults with complex needs (Liddiard, 2018, 2015, 2014a, 2014b, 2011; Taylor, 2012; 2011; Denny and Earle, 2009; Blackburn, 2002) as well as literature related to sexuality and people with cystic fibrosis (Frayman and Sawyer, 2015; Sawyer et al., 2014; 2012; Thurston, 2009). Liddiard's doctoral thesis (2011) presents an empirical exploration of 27 disabled people's lived experiences of their sexual and intimate lives, using a critical disabilities approach. Throughout, Liddiard argues that disabled people are predominantly de-gendered, de-sexualised and are often viewed within an ableist construction of disabled sexuality and thus assigned social categories of disabled sexualities, which require reconsideration.

Liddiard's thesis forms the foundation for her subsequent publications addressing sex(uality), disability and sexual citizenship (2018; 2015; 2014a; 2014b). Liddiard (2018) shows that heteronormative discourse has very complicated and contradictory implications for both disabled women and men. Her research illustrates the often complex, invisible work performed by disabled people through participants assuming separate and interchangeable roles as teachers, mediators, educators, negotiators and managers in their sexual, intimate lives (Liddiard, 2011). She argues that the majority of participants experience emotional disablism as part of their intimate and sexual lives, and that conceptual meanings and experiences of relationships, intimacy and sex may vary enormously (Liddiard, 2018; Liddiard and Goodley, 2015). We need to:

'think differently about the ideal, neoliberal citizen who may not equate to ideas of productive, sexual normality.' (Liddiard, 2016, p. 33)

It is recognised that sex(uality) is an increasing and important feature of holistic palliative care. Taylor's, hermeneutic study (2012) explores the meanings of sexuality and intimacy with 14 older adults with LLTCs and their partners. Both the participants and their partners experienced periods of connection, disconnection and re-connection during their intimate relationships. This suggests that intimacy and sexual expression for individuals were threatened by factors including bodily changes, the physical barriers imposed by important equipment to facilitate sex, and the reality of impending death. Some participants were able to find alternative ways of connecting with their partners, while others mourned the loss of their coupled status and relationships which were considered beyond repair. Such aspects of people's lives were rarely broached or discussed by health care professionals. Although participants were unclear what role health care professionals might have played, many couples would have liked to talk about their relationship experiences with them. Taylor (2012), also outlined recommendations for life-long education opportunities, further research and health care practice related to the experiences of older adults living with a life-limiting illness in relation to sexuality and intimacy. Taylor, 2011, also discusses the

important use of equipment and sexual aids, and how advanced preparation is crucial to minimise stultification of the overall sexual experience.

In my research addressing the sexuality of 100 young adults with spina bifida and/or hydrocephalus, I focused on their experiences of adolescence, the protection of vulnerable young adults, puberty, genetics, continence management, legal matters and ethics, (Blackburn, 2002). Like Hordern and Street (2007a, 2007b, 2007c, and 2007d) and Taylor (2012), I also highlighted the difficulties for health care and teaching staff to address intimacy and sexuality with either the young adult or their parents.

Sexuality and people with life-threatening conditions

There is an existing body of research on sexuality and cancers (Kelly and Vougioukalou, 2017; Kelly et al, 2015; Kelly, 2013, 2009). In relation to childhood cancer survivors, Hordern and Currow (2003), found that sexuality is intrinsic to a person's sense of self and is recognised as an intimate form of communication that helps relieve discomfort and lessens the threat to personhood in the face of a LTC. Like Taylor (2012), Hordern and Currow (2003) report that health care practitioners struggle to accept that people with a LTC, especially older people, continue to be sexual beings. People facing a LTC may welcome the opportunity to discuss issues of sexuality and intimacy with a trusted health professional. Hordern and Currow, 2003 recognise that practical strategies to assist health professionals to communicate effectively about sexuality and intimacy require training and support. In later research, Hordern and Street (2007b) extend their enquiries to explore why health care professionals find discussions with older adults so difficult about the sexual and intimate changes that might occur after a diagnosis of cancer and for those receiving palliative care.

Research is now beginning to focus on why health professionals find this type of patient communication so challenging. As part of their three-stage reflexive inquiry, Hordern and Street (2007a, b and c) conducted research in Australia to explore how health professionals reflect on issues of intimacy and sexuality following their patients' cancer treatment. Their findings contrast with the expectations of older adults. Their research showed that Cancer interrupted the sense of self, including how individuals experienced changes to intimate and sexual aspects of their lives, irrespective of their age, gender, culture, type of cancer or partnership status. Hordern and Street, (2007b), reveal incongruence between the way older adults and health professionals construct sexuality and intimacy. Hordern and Street (2007b) also noted that sexuality and intimacy were largely medicalised so that health care practitioners' discussions were limited to fertility, contraception, erectile and/or menopausal issues. In their research, many unchecked assumptions about sexuality were made by health care professionals, based on the adult's age, diagnosis, culture, partnership status and life course. It was often regarded as confronting to communicate openly about issues of intimacy and sexuality after cancer, particularly when the clinical setting emphasised medicalised, health professional-driven and problem-based communication (Hordern and Street, 2007a).

Similarly, Forbat and Kelly (2012) recognise that the clinical environment may not always be conducive to communication about intimate matters and it may create embarrassment to discuss sex during a consultation about their cancer. As indicated in Hordern's research (2007), clinical experience and training will be required on the part of the clinician to overcome embarrassment and to explore how people address their diagnosis and their psychosexual adjustment to a new reality that may include some sexual dysfunction.

Similarly, Kelly et al (2015) in their research with couples following a man's treatment for prostate cancer noted that talking about sex and intimacy was an important topic; often requiring a senior clinician to provide the necessary gravitas it deserved. Limited

opportunities were offered to individuals or couples to talk about their psychosexual needs. This was felt to be a considerable constraint, although many individuals would like such a service. Kelly et al (2015) also note that a relational approach to couple-focused support should consider the illness experience that extends beyond the individual bio-medical model. They argue that cancer is a life-changing event that affects not just the individual, but everyone in their sphere of intimate relationships. Cancer is a condition affecting both the individual concerned and those with whom the person relates, such as partners, spouses and more casual relationships.

Throughout his research with young people and adults following cancer treatment, Kelly (2013) highlights a common theme. He argues that public embarrassment and taboo may be coupled with a lack of sexual experience and that relationships, body image, and sexuality are concerns that are defining characteristics of most adolescents in normal health, so are even more important during, and following, a cancer diagnosis and treatment which may entail prolonged periods of hospitalisation (Kelly et al, 2015).

Attitudes are often influenced by the general societal attitudes towards sexual expression by disabled people (Liddiard, 2015; Liddiard, 2014). Liddiard notes that disabled people are often:

'Presumed to lack the capabilities and capacities to embody and experience sexuality and desire, as well as the agency to love and be loved by others and build their own families, if they so choose.' (Liddiard, 2011, p. 215)

Similarly, Shuttleworth (2010) has identified a range of issues where the sex(uality) of disabled people is absent because:

'Much less investigated are the socio-political structures and cultural meanings that restrict disabled people's sexual expression and sexual opportunities, disabled people's modes of resistance and creative sexual agency in their search for sexual wellbeing, the sexual implications of the intersection of disability with identity categories such as gender, race and sexuality, the impact of different policy contexts on disability and sexuality issues, and other topics less concerned with normative functioning.' (Shuttleworth, 2010, p. 3)

Sex and relationship education (SRE)

Researchers indicate that both formal and informal sex and relationship education (SRE) are important for people with disabilities (see Frayman and Sawyer, 2015; Kelly, 2013; Liddiard, 2011). Whilst research has demonstrated that formal SRE programmes may improve knowledge and awareness, such programmes may not necessarily change behaviours or the information applied. Sarrel and Sarrel (1981, p.93) have suggested that the 'transfer of knowledge in formal settings may be likened to carrying water in a basket', suggesting that information might be lost or wasted and not necessarily absorbed or taken on board by young people. Researchers welcome the contribution that informal SRE may make to the lives of people with LLTCs (Kelly, 2013; Sawyer et al., 2014).

After the Second World War, sex hygiene became embodied in the welfare state in the UK during an era that worked to improve social inclusion and cohesion among families and to broaden access of sex information for all young people, including those with disabilities (Pilcher, 2005). The origins of mainstream SRE, particularly for non-disabled people, stem from addressing the importance of contraception, growing concerns around the rise in sexually transmitted diseases, teenage pregnancies and a desire to strengthen both the moral status of health and hygiene of the nation (Reiss, 1998).

This has been described as medical eugenics, attempting to improve the quality of the population by discouraging reproduction (Wade, 2002).

Strouse and Faber (1985) indicated in their research that 'formal' sex education programmes sometimes failed to promote responsible sexual behaviour, particularly in non-disabled teenagers. Their findings indicated that parents, as well as television, sometimes diluted the impact of school SRE. They noted that formal sex education 'rarely achieved its goals' (Strouse and Faber, 1985, p. 253).

Nevertheless, from September 2020, new guidance in England on SRE for disabled and non-disabled pupils will become compulsory in schools. Pupils will study health education, with a focus on physical and mental health well-being, with a new syllabus in relationships' education, beginning in primary school and a new SRE programme initiating in secondary school. The aim is to ensure that pupils are taught the benefits of healthier lifestyles in order to address mental resilience and well-being in personal and social relationships (DFE, 2019a). From 2020, schools will also be obliged to meet their responsibilities to enable disabled pupils to access SRE education and prepare them for adulthood (DFE, 2019b).

Relationship and socialisation processes are complex and may include content that may be better addressed outside the classroom rather than within it. The literature suggests that friends are usually an important source of sex information (see, Blackburn, 2002), whilst increasingly the media as well as written literature provide accessible alternatives. McCabe (1999) notes that people with disabilities are most likely to receive information about sex from 'other' sources, such as magazines and the internet. His study suggested that there was less discussion of sexual issues among disabled people with family (parents or siblings) and that there was no specialist sex education for people with physical disabilities, including LLTCs (The Open University with Together for Short Lives, 2016; Blackburn, 2002).

Kelly (2013) also stresses the importance of cancer education for patients and the importance of one-to-one interaction, and support with regard to sexual development and function, body image, fertility, prevention of sexually transmitted diseases, unwanted pregnancy, and romantic relationships.

For disabled young adults, their self-perception and how others respond may be influenced as much by the provision of appropriate SRE as by acknowledging their need for physical and sexual fulfilment (Blackburn, 2002). Dorner (1990) noted that young adults with spina bifida and/or hydrocephalus almost always had an interest in the opposite sex, but lacked opportunities to meet and have relationships, or to access SRE specifically related to their conditions. Liddiard (2011) noted that, for disabled young adults, discussing SRE was seen as difficult for parents; friends and peers were the first choice for such conversations.

In 1996, Shakespeare et al. reported that formal SRE in schools was often denied to disabled people and for some, this access remains unchanged (Liddiard, 2011). Despite an increasing recognition of sexual rights, research reports a reluctance to address sexuality in work environments (Kelly and Vougioukalou, 2017) and that there is a gap in specialist sex education training for care practitioners (The Open University with Together for Short Lives, 2016). Much of this reluctance can be traced to social discourses that limit the right only to talk about sex(uality) to young, heterosexual, able-bodied males and therefore deny the sexuality of those who do not fit within this model (Liddiard, 2011; Tepper, 2000).

Access to formal and informal SRE may be difficult for non-disabled people (Strouse and Faber, 1985; Edwards, 2016). Care practitioners and parents sometimes make assumptions about what information is required and what should be included, thus making it difficult and sometimes impossible to have conversations about sex (The Open University Sexuality Alliance, 2016; ACT, 2009). These views are similar to the

longitudinal research studies with young people with cystic fibrosis (see Frayman and Sawyer, 2015; Sawyer et al, 2012; Edenborough and Morton, 2010). Frayman and Sawyer (2015) in their research about young people with cystic fibrosis indicated that sexual behaviours emerged across the life- course but that these may vary according to the individual LLTC. Sawyer et al (2014) suggest that young people with cystic fibrosis may attain the developmental milestones of adolescence and early adulthood at a similar time to their able-bodied peers. Ultimately, their life chances remain profoundly affected by their disease trajectory, and have a substantial impact on their overall physical, emotional and relationship health (Gee et al, 2003).

Balancing the rights to sexual expression with ‘protection from harm’

Over the last few decades, the focus of public services on the protection of vulnerable people, whilst important, has led to an increased focus on prevention of risk rather than the enabling of rights; notably, sexual rights, and in particular, the rights to sexual expression (De Than, 2015). Society has not always acknowledged the rights of disabled people as sexual beings (Wade, 2002). Furthermore, WHO (2015) advocates, disabled people should have equivalent rights to sexual expression with only those modifications which are essential for their own and others’ safety and well-being.

Disabled children, young people and adults are often perceived as vulnerable and therefore require protection from harm (Liddiard, 2014a; Craig and Lidstone, 2012). They are sometimes seen as asexual or, conversely, hypersexual, and possibly requiring containment (Liddiard, 2014a). This has implications for how practitioners address sexuality in the work place.

Practitioners’ fears of breaking the law are often based on misunderstandings about legislation and their personal fears of potential misconduct. There is a need for greater clarity about addressing the sexuality of people with LLTCs in the work environment

(Blackburn et al, 2016). A review of the law to enable safe and appropriate sexual expression where no harm will occur is being considered so 'that everyone has the right to sexual expression, relationships and fun' (De Than, 2015, p. 86).

A President of the Family Division in England recommended that those working with and supporting disabled people should avoid:

'Wrapping them up in cotton wool... the fact is that all life involves risk... physical health and safety can sometimes be bought at too high a price in happiness and emotional welfare.' (Munby, RE MM, 2007; EWHC, 2003)

In the above case, the judges ruled that the young woman had mental capacity to give consent to sex. Not all cases have received such favourable outcomes. De Than (2015) notes that both the Courts and safeguarding agencies pay lip-service to a disabled person's right to a sex life or to sexual expression. She also argues for the individual's greater choice in taking acceptable risks whilst acknowledging the importance, at times for their need for protection.

In 2014, Evidence heard in relation to the review of England's Mental Capacity Act has highlighted a number of misunderstandings and confusion about their roles by care practitioners who felt that they were abiding with local safeguarding policies by denying individuals with disabilities their wish to spend time 'alone with a girl or boyfriend'. The most relevant Human Rights provision in current English law is The European Convention of Human Rights (ECHR), see Appendix B.

The Mental Capacity Act (2005), see Appendix B, requires that all practicable steps be taken to help a person make a decision and this includes providing information about sex to those who may lack capacity but who may wish to engage in sexual activity (Griffith and Tegnah, 2014). The right to freedom of expression under Article 10 of

the European Convention on Human Rights (see Appendix B) also requires people to be given access to information which they need in order to make informed decisions about their own lives, in this case their sexual lives and puberty.

Reproductive opportunities, choices and loss

Globally, there remain a significant number of disabled people whose needs are unmet in relation to sexual and reproductive health (SRH) (Dean et al., 2017) as well as a growing literature addressing reproductive opportunities and loss in people with physical, and sensory disabilities (Earle, 2014; Earle and Lloyd, 2012; Earle et al., 2012; Earle et al., 2008; Earle and Letherby, 2007; Earle and Church, 2004a, 2004b), and reproductive loss associated with the end of life (Komaromy et al., 2007):

‘Reproductive loss refers to experiences of miscarriage, stillbirth, and perinatal and infant death, as well as maternal death, and, more broadly, to the loss of “normal” reproductive experience such as those associated with infertility and assisted reproduction.’ (Earle et al., 2008, p. 259)

Disabled people often face discrimination and lack of autonomy with regard to their reproductive and sexual health, and consequently have limited access to reproductive and sexual health care. Sexual and reproductive health are important features of health; however, people with disabilities are not always given the resources they need to achieve this. For many, this topic is addressed in schools, at home, in doctors’ offices, or in the care of other health service professionals (Earle, Komaromy and Layne, 2012). People with disabilities are not always asked about their sexual needs. Furthermore, society assumes that the disabled people cannot or do not have issues related to making sexual or reproductive choices, particularly when a person is reaching the end of life and might want someone with them before, at the time of,

and following, their death (Taylor, 2012; Zeng, Lio and Loke, 2012; Howlett et al, 1997).

Richardson (2017) contests the significance attributed solely to 'sexual citizenship', arguing instead for the centrality and inclusion of 'reproduction' to contemporary models of citizenship. Earlier Richardson (2007) emphasises the significance of reproduction to contemporary citizenship, and argues for a more careful delineation of the relationship between sexuality, reproduction and citizenship. Richardson (2017) states that parenting rights remain a contentious area in LGBT+ inclusion, adoption or access to assisted reproductive technology. As young adults with LLTCs strive for independence and sexual citizenship, efforts need to be congruent and grounded in their important views on these topics (Blackburn, Earle and Komaromy, 2018).

The literature draws attention to the importance of gender, culture and ethnicity in understanding the meaning and significance of reproductive loss, although further research is required to understand these issues fully. However, it is also important to consider that experiences of loss, and reactions to it, may vary. While every experience is unique, the level of skills of professionals will depend on their knowledge and education, as well their ability to provide compassionate, integrated, skilled care (Earle *et al*, 2012).

2.4 *Life course theoretical perspectives*

In this second section, I provide the theoretical context and literature to my empirical investigation. It is divided into four parts. First, I explore the literature related to the life course, such as (Elder, Johnson and Crosnoe, 2003; Elder, 1985). Then, I explore the conceptual linkages with the life course principles: transition points, liminality,

uncertainty and biographical disruption, and how these relate to the sexuality within an uncertain life course of the young adult research participants.

A life course perspective, the term I use in this thesis, was initially described by Thomas and Znaniecki in *The Polish Peasant in Europe and America* (1918). Thomas and Znaniecki researched the life histories and life course trajectories of their participants, suggesting that these provided invaluable data for sociologists. Like other researchers, they argued for a longitudinal approach to life history, engaging

‘many types of individuals with regard to their experiences and various past periods of life in different situations’ and reporting ‘groups of individuals into the future, getting a continuous record of experiences as they occur’ (Volkart, 1951 (p. 593).

Their arguments and approach were the forerunners of more recent research which has shaped the development of life course theory and perspectives, such as the works of Professor Glen Elder, for example, Elder (1998, 1996, 1995, 1994, and 1985).

A life course perspective, which is sometimes referred to as life course theory or a life course approach, has been defined as the:

‘Sequence of socially defined events and roles that the individual enacts over time.’ (Giele and Elder, 1998, p. 22)

As described by Earle, 2014, a life course perspective, is a leading theoretical approach in sociology and, in particular, health research:

‘A life course approach is a multi-disciplinary area of study that makes sense of people’s experiences, their decisions and their understandings of life-events

within a framework which places these in their appropriate social, structural and cultural settings.’ (Earle, 2014, p. 151)

A life course perspective has also demonstrated that events and decisions do not only relate to the circumstances of people’s lives (Earle, 2014). Social, physical and emotional development may also contribute and impact on a young person’s experiences. I argue that a life course perspective (Elder, 1975) is well suited to underpin the exploration of the sexuality, relationships and reproductive aspirations of the young adults with LLTCs, because it:

‘assumes that there are (a) recognised stages in a person’s life (for example, childhood, youth, adulthood and later life); (b) transitions or rites of passage (for example, transition from young person to adult or student to professional) and (c) specific life events (for example, birth, marriage and death). These life stages, transitions and events are understood to be part of ‘normal’ life and this serves to structure how people understand the world and their role in it.’ (Earle, 2014, p. 151)

In particular, the life course perspective draws attention to linkages between individual lives and the historical and socioeconomic contexts in which people’s lives unfold (Elder, 1975). It assumes that lives are influenced by interconnecting features; chronological age, social and ecological structures, and historical change (Elder and Johnson, 2003). Chronologisation plays an important role within the life course and life events of people with LLTCs, for instance the impact of receiving information about having a LLTC may differ depending on when this information is received and on the individual’s age, see Chapter 4.

Over time, Elder developed his life course theory (Elder, 1996; 1995; 1994; 1985). His approach encompasses the themes of liminality, transition theories and rites of

passage, biographical disruption and uncertainty of relevance to my research. These themes also represent different but compatible perspectives along the life course in relation to intersectionality; 'the relationships among multiple dimensions and modalities of social relationships and subject formations' (McCall, 2005).

Van Gennep ([1909]1960) demonstrated the relevance and efficacy of transition points and 'rites of passage'. Bury's (1982) concept of biographical disruption related to crises or discontinuities in the life course, Turner's (1969) theme of the unsettling influence of liminality. Penrod (2007) highlights the feeling of uncertainty in people with cancer. To inform my thinking in this research and to prepare the research questions and interviews, I have employed the model in Figure 2 to show how these perspectives work in synergy together. Rather than adopting one perspective, I move between them to form a bigger picture within the life course and how these relate to the young adults in this research. The model was used primarily to assist me with the literature review and the development of the research design.

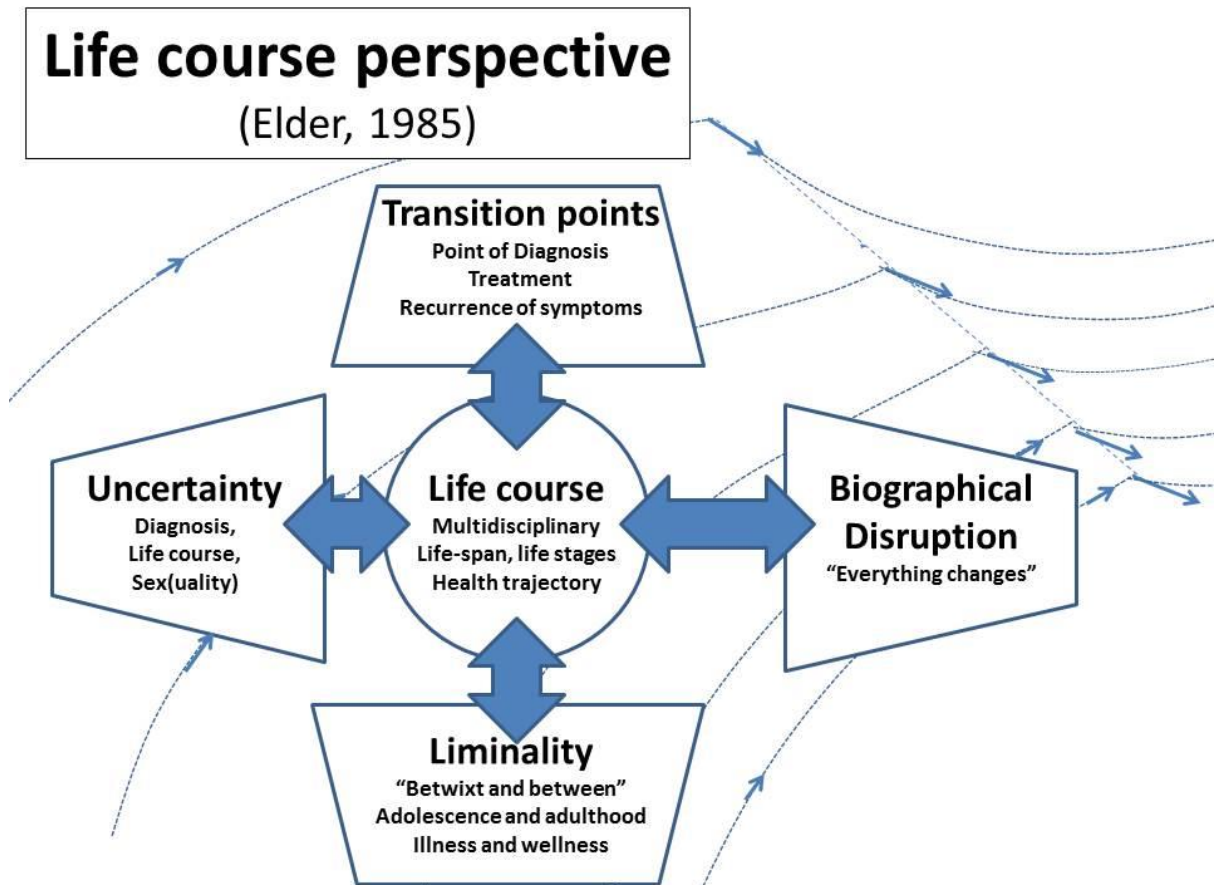


Figure 2: My conceptual model for applying Elder’s ‘Life course Perspective’ to the research design and data analysis. The model emphasises that the four “surrounding” concepts – transition points, liminality, uncertainty, and biographical disruption. These are aspects of the same holistic perspective, the life course perspective.

Whilst events do not necessarily follow sequentially, they constitute the totality of an individual’s experiences, moving from childhood to adolescence, adolescence to adulthood, wellness to illness (Giele and Elder, 1998). There are nuanced differences between life span and life course. Life span refers to the duration of life and characteristics that are closely related to age but do not necessarily cut across time and place. A life course perspective elaborates the importance of time, context, process and meaning on human development and family life. The family is perceived

as a small social group within a major social context, (Elder, Johnson and Crosnoe, 2003).

Aging and developmental changes are ongoing processes that are experienced throughout life. As such, the life course may reflect the intersectionality of social and historical factors with personal biography and development in which the study of family life, individuals and social change may ensue (Hareven, 1996; Elder, 1985). For instance, parents may have difficulty in relinquishing their caring responsibilities if their child has an uncertain life course and this may prove problematic (Jordan, Price and Prior, 2015).

Denny stresses the importance of an holistic approach:

‘a person’s whole life, to better comprehend their present experiences and beliefs.’ (Denny, 2016, p. 162)

Giele and Elder (1998) used a time-focused approach to explore the complexities of individuals’ lives using both longitudinal and non-longitudinal sociological approaches. They found that the interrelationship between social structures and the impact of time, history and location sometimes influenced individuals’ lives.

Life course research is also distinctive in weaving a ‘fabric of methodological pluralism’ from the social sciences and humanities (O’Rand, 1996, p. 53). Continuity and change, social structures, and the relationships across time, place and lives as contexts for developmental processes are unique elements of life course research (Elder and Johnson, 2003; Settersten et al, 2005; Elder, 1996). Over the last three decades, life course researchers have acknowledged the importance of the physical body (Elder, 1996) as well as the mind, recognising that the body and mind are frequently inseparable but inextricably linked (Magnusson and Torestad, 1993). This important

paradigmatic development in life course studies resonates with nursing research, policy and practice, focusing on the physical and psychosocial responses and requirements of individuals, over time relating to their health and illnesses (Black, Holditch-Davis and Miles, 2009). This is in common with the conceptual framing of my thesis.

Life course: Principles

A fundamental assumption of the life course perspective is that lives are lived in a reasonably ordered manner and may be shaped by age, social structures and historical change (Elder and Johnson, 2003). Life course draws on several distinct principles: time and place, and life-span development; timing; agency; and linked or linking lives over space and time (Black et al, 2009). I explore each of these components in turn.

Time, place and life-span development

Human lives are shaped by questions of when and where, in a socio-historical sense, making the principle of time and place and important in life course research (Elder, 1996). This is particularly significant for the young adults in this research where life course may be uncertain. The other principle, life-span development, is characterised by the view that humans develop in biologically, socially and psychologically meaningful ways beyond childhood (Elder, Johnson and Crosnoe, 2003). New situations encountered in adulthood are sometimes shaped by earlier experiences in childhood and their attached meanings (for example, Marshall and Mueller, 2003); suggesting that how parents support and care for a young person with a LLTC may be influenced by their previous experiences of illness with other family members or friends.

Timing

The concept of time in this thesis refers to the chronological ordering, rather than situating events in historical cultural contexts. Although life events are not predetermined, if the order changes, physical and social consequences may vary from those that occur within the expected timing (Elder, 1994). Similarly, certain LLTCs manifest at different stages of childhood and adolescence. This is a biological event that may not have been pre-determined, unless a diagnosis was confirmed before birth. Equally, the late onset of diagnosis of a LLTC may be a pivotal moment in the young person's life, but the individual's physical and emotional development and support from those close to them may influence and shape their responses.

Linked lives

'Linked lives refers to an integration of social relationships extending beyond formal family ties, such as friends, neighbours, and work colleagues who provide a "distinct orienting context".' (Marshall and Mueller, 2003, p. 11)

Social linkages influence how individuals interpret life events (Marshall and Mueller, 2003). People vary in the manner and degree in which they integrate social norms, relationships and institutions. Integration may be disrupted under certain circumstances (Giele and Elder, 1998); influenced, for example by the deterioration in the individual's LLTC. The lives of young adults with LLTCs and their parents are shaped by their practical care and medical treatments and these may bring with them social linkages with their carers and supporters.

Transition points

Van Gennep ([1909] 1960) identified a typical pattern of 'rites of passage', transition points, see Figure 2. Van Gennep's theory describes the way people move through life in three distinct phases (Kralik, Visentin and Van Loon, 2006). Firstly, pre-liminal rites (rites of separation) are characterized by removal of the individual from their 'normal' social life, which may dismantle institutional identity (Van Gennep, 1960). Secondly, liminal rites (rites of transition) refer to customs and rituals of the individual when they are in a liminal state, perhaps isolated or excluded, in a state of 'limbo' or, as (Draper 2003, p. 63) notes, 'in no man's land'. Finally, post liminal rites (rites of incorporation) occur where the individual is re-instated into society and adopts and creates a recognised or a new identity and/or status. Van Gennep's three-phase approach to transition continues to influence current transition thinking (Kralik, Visentin and Van Loon, 2006). Van Gennep's work was further developed by Turner (1969), indicating the way that 'rites of passage' throughout the stages of human life are marked by socio-cultural rituals. Martin-McDonald and Biernoff (2002, p. 347) state

'that rites of passage occur when there is a transition in cultural expectations, social roles, and status and/or condition or position, interpersonal relations, and developmental or situational changes to being in the world'.

Transitions are also seen as entry points for new periods or roles within life course trajectories (Hagestad, 2003). In this research, I suggest that transition is a gradual change, often associated with acquiring or relinquishing a role, for example, parenting a child or young person with a LLTC, and the additional responsibilities for parents or carers that this requires, as well as accepting the young adult's desire for independence when ongoing care and support may still be required in adulthood (Jordan, Price and Prior, 2015). Moving from school to employment or further

education, from adolescence to adulthood, and from cure to recurrence of a LLTC within the illness trajectory are recognised as transition points and different phases in people's lives (Elder and Johnson, 2003). Within the life course, individuals may occupy various roles within social institutions, such as a mother within a family (MacMillan and Copher, 2005), or a young adult with a LLTC still living with parents (Beresford, 2004; Thomas, Bax and Smyth, 1989). Transition has also been used to describe a woman's developmental challenges in becoming a mother (for example, Kitzinger and Kitzinger, 2007), and in perinatal research related to cystic fibrosis (Frayman and Sawyer, 2015). In the same way, transition theories relate to fathers and fatherhood (Draper 2003, 2002, 2000).

Transition sits on the initial and final threshold of a physical or psychological state and is therefore liminal. The key feature of that liminal phase is that structural rules and norms are suspended (Turner, 1974). Transitions are entry points for new periods or roles within the trajectories (Hagestad, 2003). For the purpose of this thesis, transition means moving from adolescence to adulthood within the illness trajectory (Elder and Johnson, 2003), from wellness to illness and from life to uncertainty around death.

Thus turning or transition points involve both substantial and sometimes abrupt change(s) from one state to another (Cairns and Rodkin, 1998). Such a transition point may include the recurrence of a condition following treatment for people with leukaemia or a benign brain tumour, or for adults with breast cancer (Trusson, Pilnick and Roy, 2016), those with other cancers (Kelly, 2013; 2012 and 2009), or other chronic illnesses (Balmer, Griffiths and Dunn, 2015; Bury, 1982).

There are other terms that are often synonymous with transition. Transition is linked to the notion of self -identity and how it is affected by disruption (Kralik, 2002; Young et al, 2002).

Transition is an adaptation to change, rather than returning to a pre-existing state (Kralik, Visentin and Van Loon, 2006). Transitional processes require time as people gradually disengage from old behaviours and ways of defining self. Nurses working alongside people can help them identify changes forced by illness and seek new possibilities from disruptive experiences (Kralik, 2002; Kralik et al, 1997). Transition entails change and adaptation, in areas such as developmental (for example, child to adolescent, adolescent to adult, for example, personal, and relationships situations. Common to these experiences, is the dislocation, disorientation and disruption caused to the individual's life and their need to find new ways of living in a world that enables positive changes to happen (Kralik, Visentin and Van Loon, 2006).

Liminality

Liminality derives from the Latin 'limen' meaning margin or threshold margin.

Liminality means:

'neither here nor there, betwixt and between all fixed points of classification.'
(Turner, 1969, p. 232)

'A limen is not just a line or a boundary serving as limit between one space or time and another, but a sensitive threshold which mediates transformation as one form-of-process becomes another. Any event of becoming presupposes the creation and operation of such delicate and volatile tipping or turning points.'
(Stenner, Greco and Motzkau, 2017, p. 142)

As noted in the previous section, the liminal self was first conceptualised by the anthropologist Van Gennep (1909 [1960]), whose seminal text on 'rites of passage' defined three periods of liminality. Van Gennep classified the phases of liminality as preliminal (rites of separation), liminal (rites of transition) and post liminal (rites of

reincorporation). Liminality has since been used as a conceptual framework to explore the rites of passage through transition in illnesses such as cancer (Blows *et al*, 2012; Navon and Amira, 2004). However, this has mainly been used as a framework to explore and describe the processes of transitioning, rather than a deep exploration of the conceptualisation of the self in chronic illness. The concept of liminality may be useful in helping to understand this period where individuals feel in limbo, as they may experience some symptoms, receive information or a diagnosis, but the full picture and impact of what this means for the individual is yet to emerge (Strickland, Worth and Kennedy, 2017).

There is a period of time for people, for example, with multiple sclerosis where symptoms are experienced prior to their confirmed diagnosis. This is described as the journey towards the moment of diagnosis and participants describe the events that lead up to diagnosis (Strickland, Worth and Kennedy, 2017), along with the details of the diagnostic consultation itself. For some participants in the same study, this time extended over many years, from a first episode of symptoms which had not been given a conclusive diagnosis to other participants who had received a diagnosis within a few weeks of their first episode of symptoms (Strickland, Worth and Kennedy, 2017).

Liminal phenomena may sometimes evoke negative feelings, reflecting ambiguity that threatens and unsettles a sense of order or organisation (Turner, 1969) within individuals or those supporting them. Turner describes a stage of liminality as one where the usual order of things is suspended, the past is momentarily negated and the future has yet to begin. I have included liminality as a conceptual basis on which to problematise the links and boundaries between the transition points, biographical disruption along the uncertain life course of people with LLTCs.

Trusson, Pilnick and Roy (2016) note that several studies have explored the concept of liminality in understanding the post-cancer experience but few studies have

considered biographical disruption several years following treatment. They argue that considering the on-going biographical disruption on the individual's body, relationships, identities over a longer period (in their study, 29 years), provided a greater understanding of the range of experiences young people may face between periods of illness and health and the impact on their deteriorating ill health and altered bodies (Trusson, Pilnick and Roy, 2016 and this links to biographical disruption.

The term 'liminal hotspots' have been used to describe transformational and emotional events impacting on people's lives); linking theoretical concepts of liminality to affectivity in psychology, medicine, law and the sociology of medically unexplained symptoms (Stenner, Greco and Motzkau, 2017). They describe

'a liminal hotspot as an occasion of sustained uncertainty, ambivalence, and tension in which people feel "caught suspended" in the limbo of an in-between phase of transition.' (Stenner, Greco and Motzkau, 2017, pp. 141–142)

For instance, a young adult in this research, who thinks their LLTC (for example a brain tumour) has been successfully treated, lives with the uncertainty and fear of its recurrence and so, in a sense, remains suspended in transition. The young adult's sense of order for both 'staged' and 'unstaged' events is often fragile, easily unsettled and can result in the interference of the latter (Stenner et al, 2017).

With supportive structures in place, this kind of turbulence of unstaged liminality (being ill) can be transformed into more manageable staged liminality (getting better or making a recovery, albeit temporary). In the absence of these supportive structures, liminality may become destructive (Stenner et al., 2017).

Similarly, following diagnosis of cystic fibrosis, or facing the recurrence of a LLTC, such as cancer, young adults (and their carers) may find themselves in a liminal state as was

seen in a study of eight young adults with cystic fibrosis (Thurston, 2009). The young adults' sense of identity as well as their futures were destabilised and became increasingly uncertain. The order and manner of their everyday lives was interrupted and both young people and their families found themselves living in limbo (Jordan, Price and Prior, 2015; Turner, 1969).

I have included liminality as a conceptual basis in this thesis in which to problematise the boundaries between transition points, uncertainty, and biographical disruption along the life course of people with LLTCs. Throughout this thesis, I highlight the intertwined nature of concepts and emotions that young adults and their supporters (parents and carers) use to understand their own roles within a liminal space and how this may impact on the young adults' sex(uality) and uncertain future.

Uncertainty

Uncertainty is more applicable to chronic rather than acute illnesses (Denny and Earle, 2009). Living with uncertainty is:

'a dynamic state in which there is a perception of being unable to assign probabilities for outcomes that prompts a discomfoting, uneasy sensation that may be affective (reduced or escalated through cognitive, emotive or behavioural reactions, or by the passage of time and changes in the perception of circumstances.' (Penrod, 2007, online)

Earle (2014, p.132), for instance, points out that 'the unexpected' may happen to any of us and that we all live with a level of uncertainty'. Those with LLTCs, however, live with uncertainty; they are constantly aware, even anxious, that their future is uncertain, and are trying to learn how to manage this.

Such features may link to the individual's search for and confirmation of a diagnosis, or explanation to the young adult with a LLTC about what's happening to them, with the ultimate goal of finding a treatment (Robinson, 1988). Confirmation of diagnosis in early years involves a re-construction for both parents and children with LLTCs (Jordan, Price and Prior, 2015; Karlsson et al, 2014) and for those where diagnosis remains without a label (Fraser et al, 2015; Rare Diseases UK, 2017).

There are other uncertainties, for example, how long people will live, whether their condition will recur, the nature of death (Denny and Earle, 2009), and central to this thesis, the implications for sexuality and reproduction (Earle, Komaromy and Layne, 2012). Charmaz (1999) focuses on uncertainty as a major feature of chronic illness, in its development, progression and treatment. Charmaz (2017) argues that the confusion of uncertainty begins with the diagnosis of a chronic illness, followed by the subsequent loss of what was previously an assumed continuity of life. The trajectory of the LLTC across the life course 'is not only uncertain but it is one that can lead to considerable pain and disability' (Denny and Earle, 2009, p. 8).

Uncertainties may also relate to distinct questions which arise for many people following a diagnosis, not exclusively to those with a LLTC: firstly, the problem of cause, when people ask themselves the question 'Why me and why now?'; secondly, the difficulties associated with maintaining normal social functioning, associated with the question of 'what should I do now?'; thirdly, the uncertainty about prognosis and what the future will offer, so 'what will happen to me now? These questions are explored in Chapters 4 to 7. The time leading up to and immediately following the diagnosis of a LLTC has been identified as a period shrouded by uncertainty and where individuals have an increased desire to obtain accurate information and advice about their condition (Strickland, Worth and Kennedy, 2017). The diagnosis of a LLTC changes the way one views one's self which has consequences for both biographical construction and biographical disruption, the latter which I now address.

Biographical disruption

Receiving a diagnosis of a chronic condition is widely acknowledged as a significant life changing event which may cause stress for the individual and their family (Bury 1982) and which centre on the importance of restructuring meaning during severe illness. 'Biographical disruption relates to the influence that chronic illness and disabilities may have on an individual's achievements and life expectations and how these may impact on social and personal relationships' (Bury, 1997, p. 124).

Biographical disruption is used by some authors to describe the changes to self-identity that require redefinition in the face of adversity (Gravelle, 1997; Boeijea et al, 2002; Young et al, 2002). The illness trajectory is another term that may be used to describe this transition. For example, Gravelle (1997) describes the trajectory that parents caring for a child with a progressive illness travel, raising similar issues to the transition literature, namely, living with loss and adversity, acceptance, tolerance, resilience and normalisation (Kralik, Visentin and Van Loon, 2006).

As stated in the section on *Transition*, the literature notes a striking similarity between those describing biographical disruption to those describing transition point in their lives. Whilst Young et al (2002) make reference to transition, Boeijea et al (2002) focus solely on the notion of biographical disruption, arising from the work of Bury (1982). The sociological experience of ill-health as chronic illness is a feature that became notable in the late twentieth century in Britain, largely as a result of a significant decline in mortality from infections, and the increasing number of people, defying life expectancy and living longer (Fraser et al. 2014; Beresford and Stuttard, 2014).

Biographical disruption means the loss of self-worth initiated by the onset of a long-term illness or a condition creating a new sense of 'normal', and irrevocable changes in the individual's life. Bury's research on biography and the life course (1991,1982) is

highly relevant in so far as he demonstrated how chronic illness creates 'biographical disruption', linked to uncertainty, across a number of transition points in people's lives.

Bury is concerned with how the 'meanings' of our everyday encounters can change significantly with the onset of a chronic illness, as specific aspects of the condition present over time and not necessarily within a defined period. Bury also sees the experience of living with a chronic illness and disability as having the effect of cutting across societal beliefs, the specific meanings attached to chronic illness and disability strongly influence the societal expectations of what an individual is able to achieve or not, both in terms of the problems, social costs and consequences of a condition such as rheumatoid arthritis and the (symbolic) significance or connotations that particular illnesses carry. Bury, (1982) describes how this leads to a loss of confidence in the body, and thus a loss of confidence in social interaction or self-identity. This concept brings into focus the meaning of illness for the individual, as well as the settings in which it occurs.

Bury (1991) also highlights the potential for an 'active coping' response to chronic illness, utilising Corbin and Strauss's (1991) notion of 'comeback'. Corbin and Strauss describe this notion in two ways: the 'physical' which refers to the activity by the person when undergoing medical treatment and rehabilitation; and the 'biographical', which is the attempt by the person to reconnect their present and future with life prior to their diagnosis. Bury, 1981, also employs the term 'coping' in terms of different kinds of adaptation rather than the normative use of 'successful' or 'unsuccessful' responses of living with a chronic illness. Both Bury (1991) and Corbin and Strauss (1991) look beyond the factors associated with a biomedical understanding of the LLTC, which typically may focus on the mechanics of functional limitations and restriction in activities caused by the specific LLTC.

Living with a chronic illness is seen as a struggle in which the individual tries to maintain 'normality' in the face of the uncertainty associated with degenerative, LLTCs, as Kelly et al (2015) note in their research related to sexual recovery following cancer treatment. The success of finding answers or an explanation for a LLTC may compound, mitigate or legitimise biographical disruption (Bury, 1991, 1982).

Similarly, young people often experienced biographical disruption, not only at the point of diagnosis, but also when they have temporarily escape from being sick (Parsons, 1951) and enter an interim phase between:

'being ill and feeling better'; 'a neither here nor there between all fixed points of classification' (Turner, 1969: p.232).

This is a phase which is characteristic and not uncommon to many people with LLTCs.

2.5 Conclusion

I have contextualised this review within current knowledge of the empirical literature specifically related to sexuality and disability, focusing on adults with LLTCs. This review has highlighted that there is still a paucity of literature specifically related to the sexuality of young adults with various LLTCs. There has been a resistance by both the public and professionals to address sex(uality) within an uncertain life course and this is indeed an emergent issue meriting exploration (Kelly and Vougioukalou, 2017). There is also evidence of avoidance of discussion by care practitioners, concerns about 'protection from harm', and possibly a lack of expertise, understanding and confidence to talk about specific issues related to sex(uality), particularly as people transition from childhood into adulthood, from wellness to illness, but who are unlikely to reach old age.

In drawing the literature to a close, I argue that young adults with LLCs transitioning to adulthood are a new 'liminal group', located somewhere in-between (Turner, 1969) children's and adult health and social care services, but with limited provision (Morgan, Blackburn and Bax, 1995). They are a growing population with specific requirements across an uncertain journey along their life course.

The young adults who are the focus of this research are set apart from birth by their life-limiting or life-threatening conditions. This separation is through the symptoms and treatment(s) of their condition(s), which usually create long-term dependency upon others into adulthood (Ledger, Shufflebotham and Walmsley, 2016). There is also an impact through the limitations the conditions placed upon their quality of life and uncertain life course and the ways in which these adversely affect their future independence, hopes, ambitions and aspirations (Abbott, 2015). My discussion of the theoretical concepts of transition points, liminality, uncertainty, and biographical disruption has foregrounded them as aspects of my overarching conceptual model, outlined in Figure 2; Elder's 'Life course Perspective'.

Chapter 3 Methodology and Methods

Introduction

This chapter is divided into four sections where I explain the key theoretical and methodological standpoints, the choices and research methods, some of the challenges and the reflection process relevant to this thesis.

First, I address the approach within the qualitative methodological paradigm, explaining the strategic process and design that has influenced my methodological model and choices and how they have informed my philosophical stance in planning and executing this research.

Second, I explain why I selected certain methods, the procedures and research techniques I used and how they relate to my methodological position. I go on to discuss how I implemented them and describe such checks that are necessary to help establish their credibility and the authority of my findings.

Third, I explore some of the challenges of undertaking 'sensitive research' and the ethical issues I encountered in doing so, drawing particularly on, for example, and Jepson, Abbott, and Hastie (2015), Law and Urry (2003), and Lee (1993).

Fourth, I reflect on my methodological approach and emphasise how carrying out qualitative research is a process requiring reflection on the ethical boundaries of the interview encounter. In particular, I explain how I managed my own subjectivity through maintaining a reflexive approach, turning primarily to Barbour's ideas on reflexivity and the way the research is 'framed and carried out' (Barbour, 2014, p. 12).

3.1 *Methodology: Choosing a pragmatic qualitative approach*

Crotty (1998) distinguishes between different frameworks on the basis of epistemology, theoretical perspective, methodology and methods. Similarly, Gadamer (1989), argues that before deciding on a method, researchers require a philosophical understanding of epistemological and theoretical perspectives to enable selection of the right method to facilitate appropriate data interpretation and analysis. I have adopted Crotty's approach as I believe that it provides a helpful structure to clarify and conceptualise the foundation for this research, how my ideas underlie and fit together, and deliver the necessary intellectual rigour.

My research effort is located within the epistemological framework of social constructionism with an interpretive theoretical perspective which acknowledges that individuals and groups construct or conceptualise their own versions of reality (Barbour, 2014; Liddiard, 2011; Gillespie-Sells, 2001; Crotty, 1998), contingent on the historical, locational and cultural influences that people experience (Crotty, 1998). Both the participants and I, as a researcher, bring individual narratives and histories to the research, embedding these within the wider history and culture to which we both belong (Crotty, 1998).

Social constructionism emerged partly because of dissatisfaction with the positivist epistemological approach which ignored the relevance and meaning of people's lived experiences (Crotty, 1998). Social constructionism seems to me to be consistent with a more socially inclusive approach, and therefore relevant to engaging in research about the sexuality of people with LLTCs.

Interpretivism is based on humanistic philosophy and provides a wide basis for qualitative research, tapping into different types of curiosities that help to pursue and

analyse data straddling different paradigms; while playing to the different strengths, skills and individual experiences of the researcher (Barbour, 2014).

Qualitative methodology

Within this framework, I adopt a broadly qualitative approach, concurring with Barbour (2014) that a qualitative approach provides a deep and meaningful explanation of the participants' data 'within the real world' (Barbour, 2014, p. 9), which for me was the real world of young people with LLTCs and what lies behind their stories, the 'why' and the 'what'.

This allows me to explore how sex(uality) is defined and experienced by young adults with LLTCs, how it is interpreted by their supporters, both professional and non-professional, as socially constructed phenomena, with the potential to generate new perspectives, drawing on the empirical findings in this research.(Barbour, 2014).

A qualitative approach aims to capture the experience and views of all four groups of participants in this research, (described in 3.2) and to elucidate how this might influence the thoughts, feelings and practices of their lives (Silverman, 2005; Charmaz, 2004). I believe it is effective in exploring a range of attitudes, experiences and meanings across different research contexts and disciplines whilst focusing on people's everyday lives and situations (Barbour, 2014, 2013; Silverman, 2013; Barton, 2005; Crotty, 1998).

Pragmatic

I have elected to follow more pragmatic and 'hybrid traditions' as described by (Barbour, 2014, p. 40). This is because of the range and complexity of the participants' conditions that require the consideration of 'strategies and techniques for analysing

how competing discourses are articulated in social settings.’ (Miller, 1997b, pp. 164–165). Additionally, my approach is informed by the theories of life course perspective.

The Life course perspective

I have chosen a life course perspective for this research as I believe it helps to demystify and present a number of experiences of people, particularly participants with chronic illness (Liddiard, 2018, 2011; Barbour, 2014; Beresford and Stuttard, 2014; Beresford, 2004). It also focuses on the influences that shape an individual’s pathway from conception to birth through childhood to adult life, as well as for those who maybe living in a perpetually liminal state, ‘neither here nor there’, ‘betwixt and between all fixed points of classification’ (Turner, 1969, p. 232) or who have reached a specific ‘transition point’ on their illness trajectory (Van Gennep, [1909 [1960]). I argue in this thesis that people with LLTCs defy classification for two reasons, first, because they were not expected to reach adulthood; second because, having reached adulthood in terms of chronological age, they are likely to have an uncertain and shortened life expectancy (Earle and Blackburn, under review).

Furthermore, life course supports a multidisciplinary health and social care focus on the study of people’s lives, the social changes and structural contexts (Denny, Earle and Hewison, 2016). In doing so, life course attempts to make sense of people’s experiences, their decisions and their understandings of life events (Earle, 2014) and assumes that individuals can exercise agency which, whilst constrained by a range of factors, enables individuals to make personal decisions about their lives (Giele and Elder, 1998). This assumption could apply to many individuals in every-day situations. However, for individuals living with LLTCs, their ability to make choices through the life course is influenced by a much wider range of factors. Although some limitations can be ameliorated, people with LLTCs are often constrained by the limitations of physical

and cognitive impairments. These individuals often require practical support to manage their disabilities– relying on people or physical adaptations in the home to assist them (Beresford and Oldman, 2000), thus adding an extra layer of complexity with respect to realising their sexual rights. (Earle and Blackburn, under review). In summary, I have chosen a life course perspective to inform my understanding of the views and experiences of young adults with LLTCs about sex and relationships and fits with my methodological approach.

Answering the research questions

The rationale and the aims and questions for this research were outlined in Chapter 1, Section 1.1. Here, I explore the reasons why I have selected a qualitative approach to answer them.

This thesis aimed to seek the views primarily from thirteen young adults with LLTCs, age 16-39 years in England, about sex, sexuality, relationships, intimacy and reproduction; what these meant to the individual participants, and how their views and experiences were shaped by their understandings, opportunities, personal experiences, reproductive aspirations and choices. From there, I sought the views on the same questions from ten parents, two partners and ten care practitioners.

Like Crotty (1998), I believe that it is possible to capture the reality of people's lives through face-to-face interviews in a meaningful and thoughtful manner, so this is why I chose to seek answers to the following research questions.

I considered that the following questions would generate responses and facilitate both informal and professional dialogue between the participant and me, as the researcher. (See Appendix J for the interview prompts linked to the research questions).

1. What are the views and experiences of young adults with life-limiting or life-threatening conditions (LLTCs) about relationships, intimacy, sex and reproduction?
2. To what extent are young adults, over 16, with LLTCs prepared through education and information, and enabled to make relationships, and sexual and reproductive choices?
3. What are the relationship opportunities and experiences for young people with LLTCs?
4. What are the views of partners, parents, carers and care practitioners, who support people with LLTCs, on all these subjects?

The research aims, and questions were intentionally framed in a broad and open way, to highlight the fact that little has been documented about the knowledge and specific relationship, intimacy, sexuality and reproductive experiences of young adults, 16+ with different LLTCs in England. The questions explore the range of understandings and experiences about sex(uality) from all four groups of participants (young adults, partners, parents and care practitioners).

In this first section, I have argued why a qualitative approach allows me to explore how sex(uality), and its various components, is socially constructed and has the potential to generate new perspectives drawn from the empirical findings in this research (Barbour, 2014). A qualitative approach aims to capture the experiences and views of all four groups of participants, and to elucidate how this might influence the thoughts, feelings and practices of people's lives (Silverman, 2013; Charmaz, 2004).

3.2 *Methods and rationale*

Qualitative research may involve the use and collection of various empirical materials, such as case studies, personal experiences, introspective life stories, face-to-face

interviews, and observational, historical, interactional and visual texts to describe the routine and problematic moments and meanings in individuals' lives (Silverman, 2013). In this section, I discuss why I selected semi-structured, face-to-face individual, interviews in preference to other techniques. One face-to-face focus group was undertaken with a group of young adult men with LLTCs to test the research questions, before the main study, as recommended by The Open University Human Research Ethics Committee.

Choice of method: Face-to-Face Interview

Researchers have successfully used a wide range of interconnected methods in order to explore and answer specific research questions (Denzin and Lincoln, 1994). Thus I have adopted a more pragmatic orientation, employing qualitative research methods as distinct from quantitative. Although quantitative and qualitative research seek to answer different types of research questions, the first using numbers and the second focusing on the spoken word and text, the two approaches may sometimes be used in a complementary way. Some researchers may incorporate both methods together, adopting a mixed methods approach (Silverman, 2013; Barbour, 2014). I initially considered using mixed methods. I had previously used quantitative questionnaires in sexuality research with 100 young adults with spina bifida and/or hydrocephalus (Blackburn, 2002), alongside semi-structured, face-to-face interviews. In that research, the richer and more meaningful data was elucidated through face-to-face interviews with participants (Blackburn, 2002).

I chose face-to-face, semi-structured interviews as the main data collection method for all four groups of participants in the main study. I felt that the participants would be given more voice and that their interpretations and answers to my research questions would be captured, as far as possible, in the words of all participants. Their answers were likely to be expressed informally highlighting what the participants

really thought and would facilitate greater dialogue, particularly on points they wished to discuss further. I was interested in discovering the range of individual reactions and responses to my research questions, rather than pursuing a collective response on what is often regarded as a taboo subject. I was accessing both narratives and attitudes from the participants (Barbour 2007).

Reasons for not choosing a questionnaire

My decision to choose face-to-face individual interviews in preference to on line questionnaires was not made without due consideration. Questionnaires are usually offered online, with an invitation delivered by e-mail. This may allow a larger number of people to contribute to the research, leading to a larger quantity of data. This apparent advantage must be balanced by the fact that those who respond are self-selecting and may not be able to clarify information about which they are uncertain (Wilkinson and Thelwall, 2010). I recognise that there is often overlap between approaches. Having explored the various methods, I chose semi-structured, face-to-face interviews with individuals in preference to other techniques within this thesis.

Ethics

I have adopted the four ethical principles to guide this research described by Diener and Crandall (1978); these being, no intent to harm, informed consent, a right to privacy and to ensure that deception and coercion do not occur. The British Sociological Association Guidelines, (BSA, 2002) were followed, as well as those of The Open University Human Research Ethics Committee (OU HREC, 2015).

The Open University's ethical review process took about six months, including the review of my initial application and finalising amendments after their requested pilot study. The OU HREC stipulated that I undertake a pilot study (a focus group interview)

prior to progressing to the main research study, to test people's views about the questions. The pilot study resulted in little change to the overall design of the main study (see Appendix D). People with learning disabilities were not the focus of this research, a condition of The OU's HREC. I also considered that the sex(uality) of young adults with various LLTCs was an under-explored area of research and in itself was a large enough study. The OU HREC final 'favourable review' was obtained after I completed my pilot study report.

In addition, the five hospices I invited to contribute to this research also requested separate ethics review by their internal hospice ethics committees. These reviews took around six months to complete.

Before beginning fieldwork, as I was interviewing disabled young adults, some of whom were vulnerable, I obtained an enhanced check from the Disclosure and Barring Service (DBS, 2013).

Gender

Early in this research, I considered the expectations of individuals who might identify as homosexual, bisexual, transgender, cisgender or gender fluid and did my utmost to recruit people who were LGBTQ+. I considered that young people with LLTCs who do not identify as heterosexual or cisgender may face additional challenges to the ones specifically identified in this research (Abbott, Jepson and Hastie, 2016). In the main study, I recruited nine young men and four young female adults. The ten parents and ten care practitioners were all female and there was one male and one female partner.

I recognise that there was a gender imbalance between the recruitment of male and female young adult participants, as well as the parents and care practitioner

participants in this research. A higher number of young adult males volunteered to participate, see Table two. I suggest three reasons for this, first, male prevalence is higher than female prevalence in the 18-plus cognisant age group in this population (Fraser et al., 2013, 2014). Second, more young adult men volunteered to participate. Third, a number of potential female participants had moderate to severe learning difficulties so were beyond the scope of this research approved by The OU HREC. I return to discuss gender issues in Chapter 8.

When it came to partners, parents and care practitioners, there were more female participants. This was partly because of the prevalence of heterosexual relationships in my sample), but also because mothers, generally, are happier to take part in research than fathers, and because there are more female care practitioners in the field than male.

Health and social care practitioners, particularly within the hospice sector, are largely dominated by female personnel (Devanney and Bradley, 2012, 2011; Blackburn, 2010). Mothers, generally, are happier to take part in research than fathers (Dilorio, Kelley and Hockenbury-Eaton, 2013).

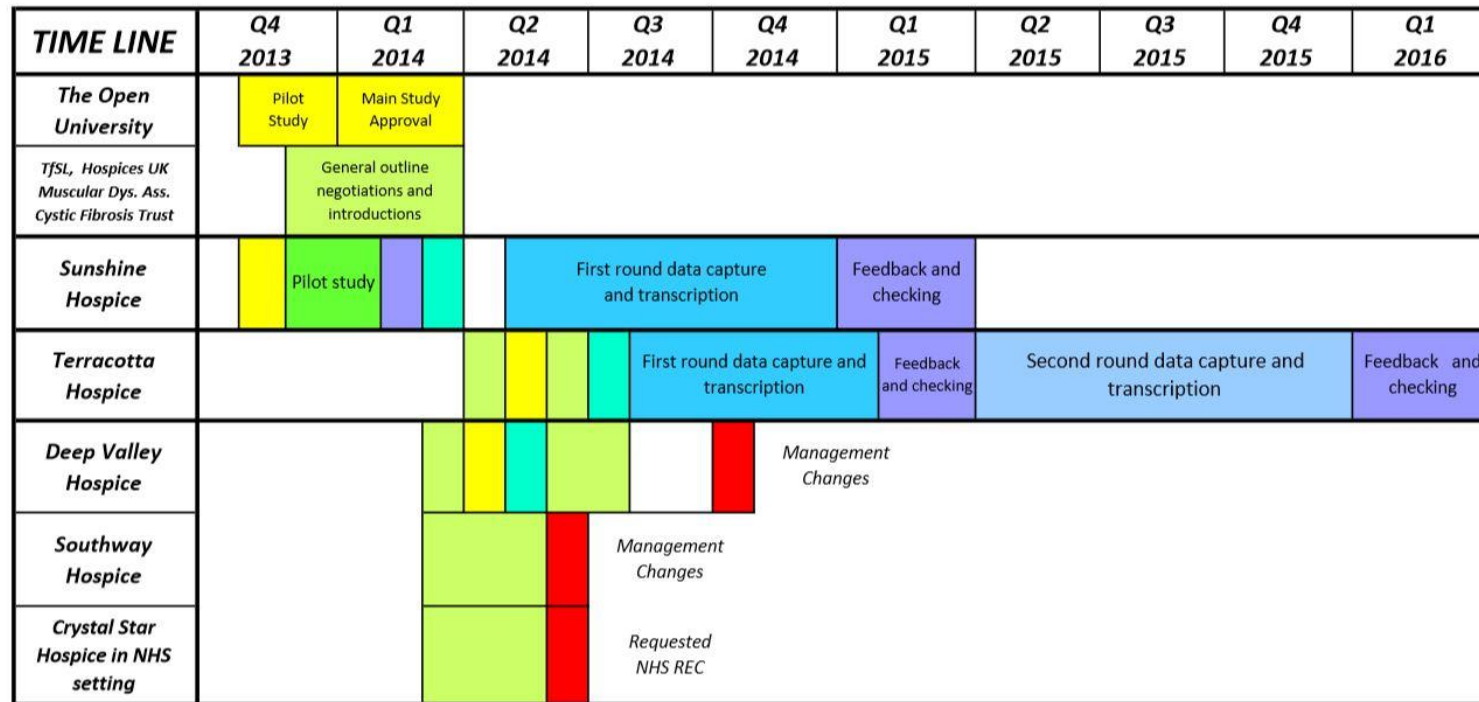
Research indicates that mothers undertake a much greater proportion of the caring responsibilities for children with disabilities than fathers (Ryan and Runswick-Cole, 2008). This has led to extensive research into the experience of mothers, but has also resulted in a lack of literature on the role of fathers. This absence of a balanced perspective on parenting children with disabilities reflects the long-standing view that fathers are less important than mothers in the work of raising children (Williams, 2014) and I return to this in Chapter 8.

Recruitment

Following The OU HREC's favourable review, the participants were recruited with assistance from national palliative care charitable organisations in the UK: Together for Short Lives, Hospice UK, thus targeting organisations which people with LLTCs were most likely to use. These charities provide advice and support to many individuals with LLTCs and their carers, and are well known within the palliative care sector throughout the UK. This process aimed to provide some distance from me as the researcher in recruiting the proposed research participants. As stated in 1.2 I had previously worked for a national palliative care charity and was known to the sector. Following discussions with both supervisors and the National Children's and Young People's Palliative Care Research Group, I focussed this research in hospice and community hospice settings in England. It was anticipated I would experience fewer recruitment difficulties, but this proved not to be the case as I discuss in Section 3.3.

In total, I contacted five children's and young adult hospices in England; *Sunshine, Terracotta, Deep Valley, South Way and Crystal Star Hospices* (all fictitious names), inviting contributions from young adults, parents and care practitioners. Following meetings with all five Hospices Boards of Trustees or their governance committees, all five hospices requested separate research ethics committee applications before granting approval for recruitment and fieldwork. Ultimately, two hospices (*Sunshine* and *Terracotta Hospices*) agreed and engaged fully with this research over two separate, six-month periods. Three hospices ultimately declined to participate and the reasons for this are discussed in Section 3.4.

Figure 3 shows the stages of the research process and a 'snapshot' of the participant interviews timeline (December 2013–March 2014, pilot study and July 2014–January 2016, main study).



Key:

- 1 HREC approval
- 2 Access negotiation and approval / **not**
- 3 Pilot study
- 4 Participant briefing
- 5 First round data capture and transcription
- 6 Second round data capture and transcription
- 7 Preliminary feedback and checking



Figure 3: Stages of the research process at each of the organisations involved in the research, including those who eventually declined, mapped onto a timeline (in quarters).

Research sample

The selection of participants is as important in qualitative as it is in quantitative research but the logistics and emphasis may differ. In this research, I sought to recruit four groups of participants (young adults with LLTCs, their partners, parents and care practitioners) from differing ages, gender, sexual orientation and cultural groups, and from different areas of England. At the outset, some clear boundaries were set; for example, ensuring that the young adults were cognisant and able to take part in this study, as advised by The Open University's Human Research Ethics Committee (HREC).

Given the nature of living with a LLTC, not all participants were able to communicate orally. Some had significant difficulties with spoken communication. Communication difficulties can impact on the ability of individuals to share their views (MacLeod, Lewis and Robertson, 2013). To facilitate the inclusion of all research participants, a range of methods were available to aid communication including the use of visual aids, such as Makaton, 2015, (which uses signs and symbols to enable communication) and other technological aids.

Some participants were supported by their parents or practitioners during the face-to-face interviews but only at the young adult's request (Earle and Blackburn, under review). Where parents and care practitioners' attended interviews, their presence may have had an impact on discussion(s). It may, for example, have limited participants discussing issues specifically related to their carers or to staff. However, in many instances, carers and members of staff who knew the participants well were very encouraging and prompted participants to talk about some difficult issues which may otherwise, not have been raised.

Marmaduke, a young, female adult had oral communication difficulties and asked both her mother, *Crystal*, and her nurse, *Spruce*, to remain at the interview. I

conducted this interview with *Marmaduke* using Makaton sex education symbolic resources (Makaton, 2015) and through her electronic computer aid, which I had been trained to use. I asked both *Crystal*, *her Mother* and *Marmaduke's* nurse to sign separate consent forms and to respect the confidentiality of *Marmaduke's* interview. In this case, I believe that their presence helped the process and did not detract from *Marmaduke's* interview contribution. In fact, their presence was particularly helpful when *Marmaduke's* Dynavox (AVdV) electronic communication aid failed and *Crystal (her Mother)* was able to help me reconnect this.

Researchers investigating young people's sex(uality) have noted that both non-disabled and disabled young people may struggle to use appropriate language in talking about sex (Robinson, 1988; Holland and Ramazanoglu, 1994; Mitchell and Wellings, 1998). I discussed language with participants in advance of the interviews to ensure people were comfortable with the terminology. So, for example, 'having sex' was used in preference to 'sexual intercourse', 'periods' in preference to menstruation.

Voluntarism

Voluntarism entails applying the principle of *fully informed* consent and thus ensuring that participants choose whether or not to take part in the research, and that exposure to risks is undertaken knowingly and voluntarily. '*Fully informed*' consent it is often impossible to achieve in practice, as researchers can not inform participants about everything (Gomm, Needham and Bullman, 2001).

Much social research necessitates obtaining the consent and co-operation of individuals who are invited to participate. Informed consent is essential whenever participants may consider taking part in research and may be exposed to substantial risks. *Consent* involves four elements: competence, voluntarism, full information and comprehension.

Competence implies that responsible, mature individuals should decide if they are given the relevant information about the research (GaFrec, 2012; British Sociology Association, 2002). It was my responsibility to ensure that individual participants were not incapable of making such decisions, either because of immaturity or moderate to severe learning difficulties.

In order to ensure that research participants had sufficient mental capacity to take part in the study, I complied with the premise of the Mental Capacity Act 2005 (see Appendix B) that everyone is presumed to have mental capacity to make decisions about their own lives, unless and until it is proved that they lack that capacity. This Act recognises that people may make unwise decisions, which does not necessarily indicate that a person lacks capacity. However, this Act does not and cannot apply to sexual matters as no one can give that form of consent on behalf of another person (The Open University with Together for Short Lives, 2016).

In cases where I questioned capacity, I invited significant others, such as parents or carers (at the choice or request of the participants) to join the young adult's interview(s), thereby minimising concerns regarding informed consent. Some young adults with minor learning disabilities asked to participate in the research, for example *Marmaduke (Young Adult Female)*. In her case, I tried to ascertain whether it would be in her best interests or not to participate, or where necessary and appropriate, to invite an advocate, carer or professional staff member to join the interviews. *Marmaduke's* mother (*Crystal*) and nurse attended the interview with her prior consent. I think it was in *Marmaduke's* best interests to have both her mother and nurse present during this particular interview, and the formalities for facilitating this were addressed earlier in this section.

Participants voluntarily agreed to participate in both the pilot and main studies and were informed that they could withdraw at any time within a pre-determined timescale. This was after completing the interviews but *before* I started my data

analysis. Participants were assured of confidentiality and anonymity and were given information on how the research evidence would subsequently be presented and stored.

Consent and Confidentiality

Informed consent from all participants was obtained either orally or, where possible, in writing (see Appendix H). Pseudonyms were used in all interviews and documentary analysis, including NVIVO 10 and excel files, in order to preserve the privacy of participants, with all names being treated as confidential (BSA, 2017; GafREC, 2012).

As some of the data was of a sensitive nature, every step was taken to ensure that confidentiality was not breached. I have taken responsibility for the safe storage and contents of the data and have done my utmost to represent the participants' views as fairly and precisely as possible (Punch, 2008). Participants will receive a copy of the final report. I believe that I have acted in such a way as to respect and preserve their dignity as human beings at all times. Where indicated, I provided opportunities for discussion and follow-up, as well as suggesting impartial support from approved counselling agencies. Thus, the conditions of the original information and consent were and have been followed.

Preserving and, where appropriate, sharing research data is now considered best practice. Arguably this has always been the case. General Data Protection Regulation (GDPR, 2018) is about strengthening participants' rights with regard to how the participants' understand how their data will be used and stored in future.

All the research data, including participant details, interview data, and field notes and data analysis have remained secure and confidential in a secure Open University

repository and have only been viewed by myself and supervisors. All data has been backed up securely into an Open University OneDrive cloud to ensure that the data cannot be accessed by unauthorised personnel.

Research briefings

I prepared a separate summary for the four different groups of participants, outlining the aims and objectives of the research, how and where they were able to participate, and my research role (see Appendices E, F and G). Prior to fieldwork, I gave a full explanation about the research to all participants, including any information on the possible consequences of the study, data storage and retention, and its future use. Three young adults, *Lamborghini*, *Morris* and *Fiat*, wanted to ‘think about it’ but subsequently agreed to take part in the pilot focus group and main study face-to-face interviews.

Pilot study focus groups

As acknowledged previously, a condition of the Open University’s HREC was that I conducted a pilot study before beginning the main study. The purpose of this was to test both the sensitivities of the research and research questions. I elected to run a focus group with six young adults with LLTCs, mainly men with Duchenne muscular dystrophy (DMD). This focus group was conducted at *Sunshine* children’s and young adults’ hospice in England. The participants agreed to contribute, following research approval by The OU HREC and their own hospice’s research ethics committee. Bar one young man, all focus group members took part in separate face-to-face interviews in the main research study, discussed in the next section.

To provide gender balance, I tried to run a similar focus group with young adult women with various LLTCs at *Terracotta Hospice*, including women with Junior

Huntington's disease, Spinal muscular atrophy and other rare LLTCs, see Appendix I. The young women declined to participate at the last moment because they said they felt shy discussing sex(uality) with a stranger. I had met the young women on a separate occasion, in advance of the planned focus group, in order to build rapport and to answer their questions.

Despite the scepticism of some researchers, focus groups have proved to be a mainstay of research into sexual behaviour and have sometimes been adopted as an approach with more vulnerable groups, particularly in potentially sensitive and difficult research (Earle and Blackburn, under review; Barbour, 2013; Barbour and Schostak, 2011).

The pilot focus group not only served to test the research design, questions and potential sensitivities, but also to assess the important relationship between the researcher and the participants in the conduct of this research. The findings did not specifically alter the research questions or overall aims of the main study. Where relevant, the pilot study data is addressed in the data chapters (please see Chapters 4 to 7).

Some researchers believe that personal information is more likely to be shared in private settings such as one-to-one interviews whereas other researchers argue that a focus group is the perfect setting to discuss personal issues including sex (see Earle and Blackburn, under review; Oliveira 2011). The pilot work and report for The OU HREC was completed within three months and I then proceeded to the main study.

Main Study: Fieldwork overview and process

Purposive selection of participants

A purposive sample, see Appendix A, should be progressive, rather than pre-planned in the way that a 'representative' sample is pre-planned in the quantitative approach. Nevertheless, it should contain a considered balance, reflecting the balance of contingent attributes of the underlying population of young adults with LLTCs who are increasing in numbers (Norman and Fraser, 2014).

In progressively selecting participants for my interviews, I sought to avoid obvious imbalances, while at the same time aiming to 'maximise diversity in the sample and facilitate comparison between accounts/perceptions of the individuals or constituencies being studied' (Barbour, 2014, p. 336). This being so, my choice of participants was influenced by the balance of availability, access and their wellness to participate. The participants invited included a representation of the main LLTCs, please see Appendix I. The resulting selection of participants evolved over a period of time (for details, see Table 2).

Main Study Interviews

As a previous quantitative researcher (Blackburn, 2002), I wanted to ensure that I obtained enough rich and meaningful data without over-burdening participants. I stopped interviewing when significant repetitions occurred, please see Chapters 4 to 7. I then judged that I had enough data to give a creditable account of the research questions I had set myself to answer. My aims at the interview were primarily to encourage the participant to talk freely and frankly around the research questions, with the assurance of anonymity. I wanted the data to be as rich as possible, and to pursue unexpected and emergent responses with the same rigour as those which had

been anticipated. By adopting a semi-structured approach, I intentionally moved away from a formalised approach (Hanson, 2013).

My primary method of data collection was to be recorded, dialogical, face-to-face, semi- structured interviews with all four groups of participants. I adopted Dennison's (2017) arguments for the use of dialogical semi-structured interviews which:

'allows me, as researcher, (1) to take responsibility for the selection of participants and (2) to manage the participant interface responsively, probing of the participant's contribution to determine its originality and quality, and whether it is made knowingly or randomly. It enables the capture of the participant's unique "logic" and "voice".' (Dennison, 2017, p. 97)

Dennison's latter point highlights the importance of the participants' contributions and engagement within the dialogue, a point close to my heart in this research.

The interview format and design of the research questions influenced how I generated data. It was important for me to balance the research relationship by listening to the participant in an uncritical way (Silverman, 2013). Having worked in the field of palliative care for many years, I recognised that my familiarity with the subject matter might influence the collection and interpretation of the data and that I needed to focus on the individual's contribution.

Over a two-year period, a total of 39 Interviews, with 35 participants were conducted with all four groups of participants in both the pilot focus group and main study (see Table 2, and Figure 3). Of the 35 participants, two young adults (*Vincent, Young Adult Male, and Jane, Young Adult Female*) were interviewed on more than one occasion because they both had partners (*Smidge, female, and Dee, male*) who wished to participate in this research. The two partners were interviewed both with and without

Vincent and Jane. As the interviews lasted between one and two hours, this was an ambitious number to complete within the study timescale. I recognise that this is a snapshot of a few LLTCs, some of which are more common and some that are very rare.

I interviewed 13 young adults with cystic fibrosis, spinal muscular atrophy or Duchenne muscular dystrophy and other rare LLTCs and carried out one face-to-face interview each with ten carers and one face-to-face interview with each of the ten care practitioners. Six young men in the main study had participated in the pilot study focus group. Arguably, the young adults in this research were a biased sample who by virtue of agreeing to take part in the research strongly indicated that they felt that there were positive reasons for addressing the topic of sex(uality) on an uncertain life course. Parents and care practitioners who supported the young adult participants were not usually invited to be interviewed because of the need to respect the young adult's confidentiality. Follow-up face-to-face interviews were undertaken with only 9 of the 13 young adults. Four young adults did not wish to take part in a second interview. The purpose of the second interview was to cross-check, validate and discuss specific points that may have been raised by carers and care professionals.

Pseudonyms and preferences

I invited all participants to explain how they wished to be referred to in this research and why. Most participants chose their own pseudonyms. Most young male adults chose 'car names' and the women, gem stones, flowers or alternative names of their choice. For consistency, when participants invited me to assist with the choice of pseudonym, I tried to respect convention, recognising that gem stones and car names might imply a gender preference or stereotypes. This was certainly not my intention but rather my wish to respect the participants' individual requests. Notably, the choice of name raised a significant ethical challenge. One individual wanted me to use her

real name and not a pseudonym. I was keen to provide consistency and assure anonymity throughout the research so avoided using real names, as most participants did *not* wish to be identified by their real name or institution. In the case of that individual, providing the real name would make that person immediately identifiable, particularly as the LLTC was often 'rare'. I explained why I needed to be consistent with *all* the participants and to use pseudonyms for everyone. The particular participant accepted my explanation and agreed to use a pseudonym.

The pseudonyms, in Chapters 4 to 7, are written in *italic* font, as are the category and gender of the participant; for example, *Lily*, female young person, is (*Lily, YAF*), *Topaz*, mother is (*Topaz, MF*) etc. Throughout this research, I refer to a participant with a LLTC as a young adult (YA), a wife, fiancée, boyfriend or girlfriend of a participant as a *Partner (P)* and I use *Mother (M)* to refer to a female consanguineous (blood-related) parent. I refer to care providers, which may also include volunteers or a family member who provides care to another (see Ball, 2009). A *Care Practitioner (CP)* refers to a remunerated health or social care practitioner, such as a doctor, nurse, family support worker, care assistant or personal assistant. If their role is a specialist one, for example, a General Practitioner's (*GP*) or clinical nurse specialist's (*CNS*) interview contribution, then the *Care Practitioner (CP)* is referred to in that role. I interviewed one medical student who was completing a student elective at *Terracotta Hospice* and she is referred to as such or as a *CP* (see Appendix A for further explanation of the pseudonyms).

The hospices invited me to provide a pseudonym for their organisation. One hospice which declined to participate in this research stated that potential participants from their organisation would prefer to use their 'real names' rather than pseudonyms. Again, doing so would have broken the codes of confidentiality of both The OU HREC and other ethical codes of practice. This ethical consideration raises a dilemma of balancing individual choice with the collective wishes and choices of participants, as

well as the researcher's role and responsibilities in relation to confidentiality and consistency.

Building rapport and other considerations

Researchers investigating young people's 'sexuality' have noted that appropriate language in talking about sex in research contexts can be challenging (Mitchell and Wellings, 1998; Holland and Ramazanoglu, 1994; Robinson, 1988

At the beginning of the interviews, I asked the young adults and parents to describe the LLTCs to me. I did this for a number of reasons. I felt that this would enable me to engage with participants on subjects about which they were more familiar, their LLTCs, while seeking to avoid the irritation of repeating their medical stories to a stranger. Over four decades, as a clinician, I have developed experience in discussing 'sensitive subjects' with research participants. In a research study that is often perceived as sensitive, asking initial questions about intimacy and relationships maybe seen as inappropriate or difficult (Liddiard, 2011) so I saw this as part of my rapport building.

During interviews, I carefully probed and targeted my questioning to gain clarification and amplification on relevant points (Bauer and Gaskill, 2000). Throughout interview prompts were used to encourage all participants to elaborate on specific themes (see Appendix J) I did not use formally scripted questions and did not necessarily apply them sequentially, preferring instead to encourage natural dialogue.

Particular care was taken to allow the participants enough time to think about the questions before responding (Bauer and Gaskill, 2000). Where necessary, I offered comfort breaks when required and in certain cases, because of their LLTC, returned at a later date, for example, to see (*Austin, and Vincent YAMs and Jane, YAF*). A challenge

of using this interview method was that recording or documenting information during interviews sometimes appeared intrusive (Walsh and Wiggins, 2003). To overcome this, I digitally recorded all the interviews with the participants' prior permission and also used a Live scribe digital pen, as a backup. This is a paper-based computing platform that includes a smart pen. Dot-paper and software applications that change the way researchers capture, use and share audio and visual information with pen and paper (Live scribe, 2018) and provides a useful backup to interview recordings. Each interview with the four groups of participants lasted between one and two hours.

Data analysis

The face-to-face interviews were digitally recorded (with permission) and then transcribed verbatim by me. Handwritten notes and memos were used in the process of data analysis. One young adult asked me not to digitally record but gave permission for me to use a Live-scribe pen.

The data were analysed using thematic analysis; a frequently used and rigorous method of data analysis within qualitative research (Braun and Clarke 2006). I chose to use thematic analysis which involves an analysis of the text where:

'all of the talk that fits under the specific pattern is identified and placed with the corresponding pattern.' Aronson (1994, p.2).

Different themes emerge as the participants' stories are pieced together to form a comprehensive picture of their collective experience (Williams, 2014).

Table 2: All participants and classifications

<i>Participant</i>	<i>Participant type</i>	<i>Sex</i>	<i>Age Group</i>	<i>Country of Birth</i>	<i>Education Level</i>	<i>Faith</i>	<i>Household</i>	<i>Medical Condition(s)</i>	<i>Occupation</i>
<i>Alfa Romeo</i>	YA	Male	30 up to 40	UK	Special College	Christian	Lives with parent(s)	DMD	Volunteer
<i>Austin</i>	YA	Male	20 up to 30	UK	University	Christian	Lives with parent(s)	Benign recurring brain tumour	Volunteer
<i>Diamond</i>	as a Young Adult	Female	30 up to 40	UK	Mainstream College	Christian	Lives with partner	Leukaemia	Mixed
<i>Fiat</i>	YA	Male	20 up to 30	UK	Special College	Christian	Lives with parent(s)	DMD	Not working
<i>Jaguar</i>	YA	Male	20 up to 30	UK	Special School	Christian	Lives with parent(s)	DMD	Volunteer
<i>Jane</i>	YA	Female	10 up to 20	UK	Mainstream School	Other	Lives with partner	Rare life-limiting condition	Volunteer
<i>Lamborghini</i>	YA	Male	20 up to 30	UK	Special College	Christian	Lives with family	DMD	Not working
<i>Lily</i>	YA	Female	20 up to 30	UK	Mainstream School	Christian	Lives with family	Rare life-limiting condition	Volunteer
<i>Marmaduke</i>	YA	Female	20 up to 30	South Africa	Special College	Unassigned	Lives with parent(s)	Rare life-limiting condition	Special college
<i>Maserati</i>	YA	Male	20 up to 30	UK	University	Unassigned	Lives with parent(s)	Brain tumour- cancer	Volunteer
<i>Mini</i>	YA	Male	20 up to 30	UK	Special College	Christian	shared parent scheme	DMD	Special college
<i>Morris</i>	YA	Male	20 up to 30	UK	Special College	Jewish	Lives with parent(s)	Adrenoleukodystrophy	Not working
<i>Vincent</i>	YA	Male	20 up to 30	UK	University	Jewish	Lives with partner	Cystic Fibrosis	Postgraduate Student
<i>Dee</i>	Partner	Male	40 up to 50	UK	Mainstream School	Unassigned	Lives with Jane	N/A	In full time employment
<i>Smidge</i>	Partner	Female	20 up to 30	UK	University	Christian	Lives with Vincent	N/A	In full time employment
<i>Daisy</i>	Parent	Female	30 up to 40	UK	University	Unassigned	Unassigned	N/A	In part time employment
<i>Emerald</i>	Parent	Female	40 up to 50	Jamaica	University	Episcopalian	Lives with family	N/A	In part time employment
<i>Goldie</i>	Parent	Female	30 up to 40	UK	University	Episcopalian	Lives with partner	N/A	In part time employment
<i>Liliana</i>	Parent	Female	40 up to 50	UK	Mainstream College	Christian	Unassigned	N/A	Main Carer
<i>Pear</i>	Parent	Female	40 up to 50	South Africa	Mainstream College	Christian	Lives with family	N/A	Main Carer
<i>Precious</i>	Parent	Female	30 up to 40	UK	Mainstream College	Christian	Lives with partner	Leukaemia	CNS
<i>Primrose</i>	Parent	Female	40 up to 50	UK	University	Christian	Lives with family	N/A	Care assistant
<i>Ruby</i>	Parent	Female	40 up to 50	UK	Mainstream College	Christian	Lives with family	N/A	In part time employment
<i>Diamond</i>	as a Parent	Female	30 up to 40	UK	Nursing School	Christian	Lives with family	Leukaemia	In part time employment
<i>Topaz</i>	Parent	Female	40 up to 50	UK	University	Jewish	Other residential care	N/A	Main Carer
<i>Millie</i>	Social Worker	Female	50 up to 60	UK	Mainstream College	Christian	Lives with husband	N/A	Social Worker
<i>Peach</i>	Med Student	Female	20 up to 30	UK	Medical School	Roman Catholic	Unassigned	N/A	At University
<i>Oak</i>	GP	Female	30 up to 40	UK	Medical School	Unassigned	Unassigned	N/A	Doctor
<i>Ellie</i>	GP	Female	40 up to 50	UK	Medical School	Unassigned	Unassigned	N/A	Doctor
<i>Apple</i>	Carer	Female	20 up to 30	UK	Mainstream College	Christian	Lives with partner	N/A	Main Carer
<i>Cherry</i>	Care Practitioner	Female	40 up to 50	Ireland	Nursing College	Christian	Lives with family	N/A	Full time employment
<i>Elm</i>	Care Practitioner	Female	30 up to 40	UK	Mainstream College	Unassigned	Unassigned	N/A	Youth Leader
<i>Spruce</i>	Care Practitioner	Female	40 up to 50	UK	Nursing College	Unassigned	Unassigned	N/A	CNS
<i>Walnut</i>	Care Practitioner	Female	40 up to 50	UK	Nursing College	Unassigned	Unassigned	N/A	CNS
<i>Pine</i>	Care Practitioner	Female	40 up to 50	UK	Nursing College	Unassigned	Unassigned	N/A	Main Carer
<i>Focus group</i>	YA	Male	20 up to 30	UK	Various	Various	Various	Multiple life-limiting conditions	Various

I then continued to the next stage of the thematic process which involved cross-referencing my findings with existing literature (Aronson, 1994), to see how these related to present knowledge about the sexuality of young adults with LLTCs and thus enable me to identify commonalities in themes, discussions and arguments.

Analysis of the transcripts involved identifying key words, phrases, explanations or experiences which reflected the interviewees' meanings, as well as the actions and experiences that were noted during all of the interviews. I used participants' language from the interviews to illustrate the significance that they attached to the specific meanings of their experiences.

NVIVO is a qualitative data analysis (QDA) computer software package produced by QSR International and is designed for qualitative researchers working with very rich text-based and/or multimedia. I used NVIVO 10 and 11 (Microsoft windows version) to assist data analysis. For each theme, I created files of both audio and written transcript extracts within NVIVO. This software uses a code-based system, 'offering sophisticated and flexible tools' (Silver and Lewins 2014, p.70). Given the nature of my research questions, see 3.1, I analysed the transcripts, using inductive, open- coding, adopting a data-driven approach to thematic analysis rather than one which set out to answer prescribed questions. To increase rigour, I listened to the audio recordings of each interview several times to identify common themes. I then read and re-read the transcripts for each interview before separately analysing the data, using memos to add specific notes about commonalities and differences between the interviews and the themes (Earle and Blackburn, under review,).

The codes were then compared and contrasted using a dynamic process that Lincoln and Guba describe as 'going back and forth' (Lincoln and Guba, 1985, p.342) in order

to produce a 'code list'. I devised an initial code list and used this to refine and collapse the preliminary codes into categories, for example, uncertainty about 'friendships', 'love', 'sex', 'wanting a relationship', 'wanting children' 'having children'. In my final thematic analysis, I focused on five main areas: drawing on the work of Smith (2001), who has argued such documents produce or reflect 'social facts' about people or phenomenon in different social constructs (Earle and Blackburn, under review).

The themes that emerged were pieced together to form a comprehensive picture of the young adults', partners', parents' and care practitioners' discussions. The differences and similarities between the different groups of participants' views were individually analysed. The data analysis identified five key themes: 'Uncertainty along the life course,' Sexual knowledge and sex education', ' Sexual experience', 'Meanings of friendships, relationships, intimacy and sex', 'Reproductive choices, experiences and loss'. These form the basis of my data analysis, see chapters 4-7.

My data analysis focused on one the one-to-one interviews in the main study. Most participants had rarely discussed sex and relationships with anyone. The main study interviews generated a large amount of complex data. In Chapters 4 to 7, I have prioritised the contributions which were unique, important and pivotal to an emerging group of adults with LLTCs (Beresford and Stuttard, 2014).

In Chapters 4 to 7, I refer to the key themes. These include excerpts from the transcriptions to communicate how all four groups of participants responded in 'their own words', thus providing a curation of their important contributions (Hammersley, 2015, 2014, 2013 and 2007) and recognising and articulating patterns through thematic coding. Table 3 shows the process of my data analysis. Appendix K shows an example of an interview transcript and how this was analysed.

Process of Data Analysis			
1. Method of analysis for open-ended, semi-structured interview questions	2. Data familiarisation using NVIVO	3. Initial analysis to identify key themes	4. Full thematic analysis to construct and refine the narrative
<ul style="list-style-type: none">• From data participant to NVIVO 10• Transcribed verbatim, all groups of participants' interviews.• Formatted transcriptions. Checked and exported all the formatted, verbatim transcribed word documents into NVIVO.• Exported all digital audio-recordings into NVIVO	<ul style="list-style-type: none">• Inside NVIVO read through each of the documents separately to identify common, interesting or unusual themes and patterns in each of the transcriptions.• Listened to audio-recordings on NVIVO. Created memos in NVIVO on differences and nuances between audio and transcribed interviews.• Recorded my observations of interviews and perspectives on emerging themes in separate memos and used these to initially cross-check coded, emergent themes and final themes.	<ul style="list-style-type: none">• Used NVIVO 10 and 11 to open code each transcribed document, creating a separate set of codes for each open-ended question. Created back-ups on Excel spreadsheets and exported these from NVIVO to Excel files to ensure reliable access.• Ran a word frequency analysis on each document and created colour coded tags to visualise frequently occurring words.• Compared the images and lists of frequently used words with the list of codes I had created from manually coding the data. Key themes identified	<ul style="list-style-type: none">• For each key theme, I identified a set of codes. The codes were selected to reflect the most frequently occurring themes (for example, uncertainty, meanings, experiences of relationships, access to formal and informal sex and relationship education (SRE), and reproductive experiences).• Mutuality for themes that were not frequently occurring but that I was interested in exploring (for example, links between the age and confirmation of diagnosis with emerging awareness of sexuality as an 'influence' on understandings of intimacy, sex and relationships).

Table 3: Process of Data Analysis in four stages.

3.3 *Specific ethical and methodological challenges*

Gatekeeping

There were inherent and explicit prejudices about gathering data which I had to overcome in this research, particularly in relation to engaging ‘gatekeepers’. Much has been written about the important role(s) and benefits, as well as the resistance, by gatekeepers in the research process (McFadyen and Rankin, 2016 and Bryman, 2008). Gatekeeping responsibilities may include protecting and safeguarding children and young people, ‘vulnerable adults’ and families who access their institutions (McFadyen and Rankin, 2016). It is a reasonable expectation for the gatekeeper to ensure that the researcher is meticulous in adhering to ethical principles. In this respect, the gatekeeper needs to ensure that individuals within their institutions and environments are protected from coercion and exploitation at all times (Bryman, 2008).

There are a number of terms that have been used to describe gatekeeping or gatekeepers, (McFadyen and Rankin, 2016; Berg and Lune, 2004). The term that particularly resonates in this research is Berg and Lune’s (2004) as they also indicate the power relationships of those roles:

‘Gatekeepers are individuals who have the power or influence to grant or refuse access to a field or research setting.’ (Berg and Lune, 2004, p. 23)

Whilst gatekeeping is:

‘A term referring to the adult who controls or limits researcher’s access to participants. For example, the top manager or senior executive in an organization, or the person within a group or community who makes the final

decision as to whether to allow the researcher access to undertake the research.'
(Saunders, 2006, p. 49)

Research is an increasingly important feature in health, social science and palliative care as practitioners are required to respond to changing priorities in light of evidence-based research findings (McFadyen and Rankin, 2016). Thus gatekeepers and gatekeeping have a major role in enabling researchers to gain access to potential participants and sites for research, and this study was no exception. Whilst the researcher–gatekeeper relationship may work well, at times, gatekeeping may be a stumbling block, with researchers having limited or no access to sites and participants, thus impacting on the overall outcomes and success of their research (McFadyen and Rankin, 2016).

The participating hospices who were willing to assist in the research agreed to forward information to relevant individuals inviting their participation. I also accessed practitioners and clients from the participating hospice services by providing information sheets, and online newsletters. I decided not to use social media groups and websites to post information because of the sensitive nature of the research. However, it became a struggle to engage some hospice services in research about sex(uality):

"This is a taboo subject in our hospice." (Care Practitioner)

As my research progressed, it became apparent that without an inside contact, somebody who worked within the hospice and who was willing to help me gain access to participants, recruiting people would be impossible. A more significant obstacle emerged as I began to approach hospice services and other palliative care facilities, that of my research topic – sex – and some hospices did not give me permission to access participants on account of the research topic.

This was in contrast to those hospice services I contacted that welcomed the research (*Sunshine* and *Terracotta Hospices*). I worked closely with these two hospices to ensure that participation was voluntary. I was invited to visit and presented the proposed research to both their Boards of Trustees and separately to their care practitioners. They were helpful at every stage of the process; circulating information packs and consent forms and encouraging recruitment. Another hospice (*Starshine*) also initially greeted the research enthusiastically, but a change in senior management resulted in their stating that they could not assist me at this time.

The fifth hospice (*Big Valley*) initially appeared keen to cooperate. I had several meetings at the hospice with both senior medical staff and nursing management. I also provided a training session for their care practitioners to explain the research and answer questions. I was eventually advised by the ethics and research advisor to submit an application to *Big Valley Hospice's* internal Human Research Ethics Committee, although they had received 'the favourable review' documentation from The Open University HREC (see Appendix D).

Two months after that submission, *Big Valley* ethics committee asked me to make several amendments. Following further email dialogue over the next six weeks, I had a meeting with the Hospice's research ethics administrator. Subsequently, it was agreed that the research could proceed, and the administrator would be responsible for inviting participation, acting as their gatekeeper. After some weeks, I emailed the administrator to ascertain progress. Several weeks after this request, the same administrator e mailed me stating that they could not accommodate my research request at this time.

I replied, offering to come and talk to potential participants and, following discussions with The OU HREC and my supervisors, considered revising the timeline. Sadly, this

offer was not taken up. I reflected over the obstacles I had faced and the possible causes as to this resistance.

Where research is conducted within a gatekeeper's work environment, there is the risk that dissemination of the research findings may cause embarrassment or criticism of the gatekeeper's organisation. I suggest that there was some resistance from *Deep Valley* and that some, but not all, of the care practitioners greeted the research favourably at the training session: The requirement to gain formal ethical approval through several ethics committees involved in the early negotiations also impacted on the overall timescales for the research.

Sensitive research

In this section, I reflect on the methodological peculiarities and challenges of qualitatively researching sensitive topics, particularly whilst engaging with marginalised or hard-to-reach groups, such as young adults with LLTCs. I believe that a life course approach worked well in this study to answer questions where the research topic may be 'sensitive', without seeking to problematise it (Earle and Blackburn, under review; Renzetti and Lee, 1990). Sensitive research generally encompasses a wide range of issues, undertaken across a variety of disciplines and settings, using a range of methods (Dickson-Swift, James and Liamputtong, 2008). Lee (1993) argues that individuals who pose a substantial threat to individuals involved in the study, may also include the researcher.

Renzetti and Lee (1993) acknowledge that all research has the potential to be sensitive, and so sensitivity should not be used to describe only particular groups or approaches to research. Fears over the sensitivity of research can lead researchers to find that they begin to 'edit' themselves out of their own research (Valentine, 2007). The fear of enacting harm through the intervention of the interview can mean the

researcher's body begins to disappear in the chase for elusive objectivity. The rigour of the ethics committee, though designed to carry out the important role of upholding the integrity of research, may also instil fear into the novice researcher.

Lee (1993) has acknowledged that reciprocity between the theory and the practice of research is important but such focus of inquiry and evaluation has only been acknowledged relatively recently. Renzetti and Lee (1993) also note that no research is so innocuous that 'sensitivity' is not a concern. They present extensive expositions on how to resolve the particular issues that those undertaking sensitive research may encounter and, in particular, what makes research contentious. They consider the relationship between research and issues of political and social power, the capacity of research to encroach on people's lives, and the potential implications researching sensitive topics may have for the researcher. They also examine the political and ethical issues inherent in the relationship between the researcher and the researched, and in the disclosure, data management, dissemination and publication of research; all features which resonate with this research.

Undertaking qualitative research on sensitive topics often raises a variety of ethical issues, in particular the issues faced by researchers during the conduct of qualitative research on subjects, such as death and dying, sexuality, homelessness, HIV/AIDS or cancer (Newton, 2016; Dickson-Swift, James and Liamputtong, 2008). This may be amplified when discussions include topics of a sensitive nature, such as sex(uality). I acknowledge that this research was 'sensitive', combining three of the most recognised taboo subjects for discussion: sex, end of life and death (Liddiard, 2018; 2011; ACT and Triangle, 2011).

Recruiting research participants can be particularly problematic when research focuses upon particular individuals or groups, such as those with LLTCs, or experiences which may not necessarily be endorsed by society (Browne, 2005). These individuals and

groups are often 'hidden' because openly identifying with their particular challenges may result in discrimination. Faugier and Sargeant (1997, p.472) argue that the 'more sensitive or threatening the phenomenon being researched, the more difficult sampling may be'.

The study of sexuality is a sensitive subject because there may be risks to participants (Browne, 2005). Moreover, sexual issues are often considered 'private' and outside the 'public' realm of research. Yet Bell (1997) contends that 'probably the singular most difficult aspect of researching sexual geographies is that of access' (p.417).

I argue that it may be the subject matter or the discovery that creates this deliberation. I considered the barriers and obstacles to carrying out sensitive research. In relation to this research, the following issues were noteworthy: issues about the subject, issues for the participants and for the researcher.

Issues related to the research topic and the participants

Sensitive research may include interviews about emotionally difficult subjects or deeply personal issues, interviews with 'vulnerable populations' or research that could have negative consequences for participants, thus impacting on recruitment. From the outset, this research led me to think carefully about what I was asking participants. I was aware that recruitment would take time (discussed in Section 3.2).

The criteria for participants consisted of being aged 16 or over and having a LLTC. When selecting participants, I also looked at the possible complications related to participation, not only specifically for the young adults themselves, but for their families and the care practitioners supporting them.

In the process of accessing and recruiting participants, naturally I encountered challenges. I believe this was partly due to the way sex(uality) is perceived by society (discussed in Chapters 2 and 4 to 7). Four of the hospices had set protocols to follow about research participation, such as completing a detailed application form, including the research proposal, guidance briefing and consent forms, which would then be reviewed by their internal ethics committees, in addition to The Open University's HREC. I argue that the obstacles I faced were due to the sensitivity of the research topic.

Issues related to the researcher

I encountered inconsistent and contradictory recommendations from third sector Research Ethics Committees (RECs) which were often at variance with The Open University HREC. This raised questions about who should be the ultimate decision-maker; the University or the third sector research ethics committee.

The methods deployed for respecting participants' wishes while preserving anonymity presented challenges. The assumption that it is a 'right' to respect confidentiality and anonymity was balanced with risks of disclosure and what this meant in reality; especially when some of the young people in this research wanted to be referred to by their real name. Indeed, some participants (young adults and care practitioners) challenged the over-cautious anonymisation, which they thought diluted their contributions. Inevitably the approach had to be one of uniformity and whether one person or 35 participants wished their true identity to be acknowledged, consistency and confidentiality remained imperative. Researchers are not always clear about identifying the sensitive issues because until recently, innovative solutions addressing sensitivity have been rarely explored (Ellis, 2017; 2015).

Posthumous consent

Wherever possible, it is important to discuss and record the different phases of consent and assent in advance of death, recognising that participants nearing the end of life might die before the research is completed or published. Negotiating, agreeing and respecting the views and requests of participants following their death raises interesting ethical challenges. Parents and/or carer practitioners, sometimes inappropriately, answer and act as gatekeepers on behalf of the young adult during their lives and even after death. This raises significant challenges of seeking, renewing and maintaining posthumous assent or consent with young adult participants and I return to this subject in Chapter 8.

3.4 *Personal Reflection*

It is important, before, during and after research, that the researcher shares their reflections on their own values and decision-making as they relate to methods, context, relationships, and outcomes of their research. This sharing demonstrates awareness of the implications of how the knowledge generated impacts on the social world from whence it arose (Bryman, 2008) and upon the researcher themselves.

In this section, I reflect on how doing this research has impacted on me as a researcher, and the consequences of this for research practice and knowledge generation. Notably, in this chapter, I have explored some of the sensitivities encountered when researching sex with young adults with LLTCs who may or may not be approaching death. I have examined ethical, practical and methodological issues in the context of an uncertain life course. The intersectionality between research with young people, sexuality, disability, dying and end of life highlight why certain taboos are foregrounded in this research and why I reflect that this may be the case. Some

research may be considered so sensitive that it may not be appropriate to conduct it at all. Yet, as Bourne and Robson (2013) note:

‘We have an ethical obligation to consider how our research activities might impact on our participants, yet rarely do we explore with them how they think about the research they took part in and what, if any, impact it had on their lives.’ (Bourne and Robson, 2013, p.16)

Throughout this research, I wanted to capture and express the views of others. I wanted to learn from all four groups of participants aspects about their sex(uality) that were new to me. This is the nature of dialogical learning, Dennison (2017). I was trying to explore the participants’ understandings, reporting their reactions and responses *in their own words*, as far as practicable, while simultaneously exploring my own to provide for the reader a touch-stone check on my own involvement.

The advantages and deliberations in choosing the right method meant that my research design should include the following three stages: first, progressive selection of participants (Yin, 2009, p. 31), second, engaging participants in a way that was likely to achieve an authentic and accurate account of their views using everyday language. Third, maintenance of their authentic voices through the mediation processes of selecting participants, interviewing, transcription, analysis and reporting mechanisms (Hanson, 2013, p. 395).

At each stage of my fieldwork, I strove to minimise both procedural and outcome error. In my case, dialogue with The HREC at various periods of the fieldwork to check and re-check *potential* departures from my original HREC submissions were important and I am grateful to the advice and support I received from The OU HREC throughout the research journey.

To assist my reflexivity, I made detailed notes and discussed specific issues with my supervisors. I attended clinical supervision sessions for doctoral students engaged in fieldwork, provided by The Open University. This is a model of supervision outside the realms of formal, doctoral supervision, described by Hunt, Swallow and Twycross (2014). This enables the researcher to reflect on some of the personal challenges and encounters of the research process and has helped me to reflect on aspects of the following.

The Participants

The participants' evidence did not neatly merge into a single discourse, divided into 39 interviews with four different participant types, see Table 2. However, the narratives reflected the similar, mixed, indecisive and sometimes contradictory contributions from their individual stories. In particular, some young adult participants did not wish to talk about end-of-life. In some cases, I elected not to ask these specific questions because it felt inappropriate at the time on the grounds of the young adult's ill health. Furthermore, several participants stated that they preferred to focus "on living" or to discuss other matters. Sometimes, we just ran out of time or I sensed that the participant was tired and I needed to close the interview.

I encountered inconsistent and contradictory recommendations from third sector Research Ethics Committees (RECs) which were often at variance with The Open University HREC. This raised questions about who should be the ultimate decision-maker; the University or the third sector research ethics committees.

The methods deployed for respecting participants' wishes while preserving anonymity presented some challenges. The assumption that it is a 'right' to respect confidentiality and anonymity is balanced with risks of disclosure and what this means

in reality; especially when some of the young people in this research wanted to be referred to using their real names.

3.5 Conclusion

In this chapter, I have set out the methodological approaches and methods I have adopted in this research; favouring a qualitative approach, using semi structured, face-to-face interviews. I have also explored and reflected upon some of the challenges encountered through my selected methods on some issues I encountered as a researcher; such as, gatekeeping, sensitive research and posthumous consent.

Telling and reflecting on the research story is often the only way of refining its focus, development, the literature, and the data analysis (Barbour, 2014). All prejudices and assumptions influence how we act and behave in the world and some of these features may prevent rather than assist the research process. Some of these factors may have far reaching consequences for the research and I shall return to these in chapter 8.

Thus, this chapter is not only an evolving research journey, but also a retrospective explanation of my choice of methodology, how the methods developed and why. The narratives that unfold in Chapters 4 to 7 are marked by life-stages, transitions, uncertainties and disruptive events that map out and construct how young adults with LLTCs see themselves in their world, and conversely how they see the world that they inhabit (Bruner, 1990). I now move on to explore these aspects in the first of four data chapters.

Chapter 4

Uncertainty along the life course

Introduction

‘We are an unforeseen generation, since we were not expected to still be alive and, as a consequence, organisations and agencies that were originally created to advise and support our parents/carers have been slow to respond to our needs.’
(Jon Hastie, CEO DMD Pathfinders, 2016, see www.dmdpathfinders.org.uk).

The young adults’ clinical background and the onset or impact of their diagnosis remains a defining feature of this research. As a researcher, knowledge of the young adults’ conditions and their own clinical history in relation to that diagnosis enabled me to better understand how the participants lived with a LLTC, and how it impacted upon their lives. Arguably, not to have established details of their condition(s) and the timing of their diagnosis would have been a disservice to the participants. It was my conversations with young adults or parents about the timing and diagnosis of the LLTC that made this first data chapter so significant in terms of its relationship with the four specific research questions about sex(uality) which I then explore in Chapters 5 to 7. The very nature of the young adult’s LLTC condition and, in particular, their point of diagnosis, made a major difference to the central conceptual issues of the liminality, biographical disruption and uncertainty across the various transition points across their life course, explored in Chapter 2.

This chapter shares with the reader some of that knowledge around conditions, clinical care, and the stories around how the young adults with LLTCs were first diagnosed. This first contextual chapter is a necessary preliminary laying the foundations for the following three data chapters, Chapters 5 to 7. The diagnosis and its timing were explicitly predicated on an in-depth understanding of the LLTCs and their impact on both the young adult and their supporters. This understanding was central to gaining

access to participants, planning the data collection process, analysing and interpreting that data, and collating and presenting the findings. The LLTCs and their impact on individuals were defining features of the research. This is more than scene-setting. Without this knowledge, I argue that the sex(uality) of young adults with LLTCS cannot be fully understood.

The importance of this knowledge was brought home to me during the interview process, when I began each interview by inviting participants to discuss their LLTC(s); some young adults had more than one. This was intended to establish rapport by enabling participants to talk about something reasonably neutral, as well as eliciting necessary information about the LLTC. In response to “tell me about your life-limiting and/or life-threatening condition”, participants gave information about their diagnosis, when and how it occurred, if they knew the name, and whether the LLTC had been confirmed or not. It soon became apparent that, where a diagnosis was named or existed, the moment of diagnosis was a particular transition point in the young adults’ or parents lives. As such, it may be said to form a pivotal disruption in their life course trajectory, concordant with Bury’s (1982) thinking. The overall significance of this became apparent when I tried to make sense of how participants looked at their LLTC, and whether the specific condition had either been explained or confirmed (Jordan, Price and Prior, 2015 and Karlsson et al, 2014) or still remained without a label (Fraser, 2015). The experiences of receiving and understanding their diagnosis formed a climactic transition point in their lives, something that shaped the attitudes and stories that they would live with for the rest of their lives:

“Until my diagnosis was confirmed, I felt normal, just like any other young person. Then everything changed.” (Lily, YAF)

I describe this as ‘a pivotal moment’, creating disruptions along the life course, something that shaped their attitudes and the stories, such as:

“Receiving the news that Lily had a life-limiting condition was a pivotal moment in our lives, both for me and our family.” (Liliana, MF)

Thus, this chapter and its structure emerged as a result of my first question and the responses that unfolded. I acknowledge that I did not ask all groups of participants this question (see Section 3.4). The young adults’ responses to this question partitioned them in a meaningful way. Their LLTC was central to understanding their lived experiences and how this impacted on their emerging sex(uality). The parents’ answers were also important in understanding their roles as primary supporters of people with LLTCs. The young adults and parents talked about when they had received a diagnosis; if the LLTC had been explained to them (Jordan. Price and Prior, 2015; Karlsson et al, 2014; Anderson and Clark, 1982), when and how it occurred, and whether it was confirmed or not, or so rare that the LLTC remained without a label (Rare Diseases, RDUK, 2017; Fraser et al, 2015). These pivotal ‘points’ varied depending on when the young adult or parent received information about their condition(s) and is my rationale for organising this chapter under the four following sections; first, those diagnosed with or suspected of having a LLTC at birth or in early childhood; second, those diagnosed with a LLTC in late(r) childhood or adolescence; third, those with a recurring LLTC, and fourth, those with an unnamed or a rare LLTC. At the end, I draw together the findings.

4.1 *The pivotal moment of diagnosis*

Inviting participants to discuss their LLTCs, I argue that this enabled both young adults and parents to discuss a subject about which they were familiar and prepared to discuss, as well as eliciting important findings about the LLTCs. This provided a starting point to then explore the relevance of when and if the LLTC bore any relationship to the young adults’ opportunities or lived experiences of sex(uality), which is explored in Chapters 5 to 7.

4.2 *Diagnosis at birth or in early childhood*

As I discussed in Chapter 2, the importance of the sociological constructions of childhood and adulthood have emerged and developed over time and across cultures. Childhood was previously considered to be a short period preceding adulthood (Denny, 2016). As a child moves from childhood to adolescence and ultimately to adulthood, the fundamental goal is to achieve independence with:

‘The desired state of stability and completion that constituted arrival at the journey’s end.’ (Lee, 2001, p. 7, cited in Denny, 2016, p. 164)

I have interpreted early childhood as the period from birth to eight years, including infancy, toddlerhood, and the pre- and early-school years (The Sexual Health Respect Toolkit, 2018; The Open University with Together for Short Lives, 2016). Early childhood specifically refers to the ten participants who told me that they had received a diagnosis of a LLTC before or around eight years of age but had not necessarily received a name. The findings from *Austin (YAM)* and other participants (*Morris and Maserati, YAMs, Daisy and Crystal, MFs*) feature in other sections in this chapter.

Five young adults (*Vincent, Austin, Mini, Jaguar and Lamborghini, YAMs*) and five parents (*Crystal, Topaz, Daisy, Goldie and Pear, FMs*) told me that they had received a diagnosis before their child was eight years, even if they did not know its precise name. Table 4 details the participants who reported receiving a diagnosis before eight years.

<i>Young Adult (Pseudonym)</i>	<i>Diagnosis</i>	<i>YA age at diagnosis</i>	<i>YA age at interview</i>	<i>Parent and research involvement</i>
<i>Marmaduke</i>	Complex LLTCs: mild leucodystrophy and possibly Pelizaeus Mezbacher and neuro disabilities	From birth, although normal Apgar score at birth. Mother knew of LLTC soon after birth but did not have a precise name.	23	<i>Crystal</i> (mother) interviewed together with <i>Marmaduke</i> and her nurse at her request on account of <i>Marmaduke</i> 's communication difficulties.
<i>Vincent</i>	Cystic Fibrosis	Around 2 years Both Vincent and his mother were informed at the same time, although Vincent did not understand his diagnosis until later.	27	<i>Topaz</i> (mother) interviewed separately from <i>Vincent</i> at his request.
<i>Austin</i>	Recurring benign brain tumours (juvenile pilocytic astrocytoma (JPA))	Age 2 and recurring benign tumours at 9, 13, 24 years.	29	<i>Pear</i> (mother) interviewed separately from <i>Austin</i> at his request.
<i>Mini</i>	DMD	Around? 5 years.	23	Parent not interviewed.
<i>Jaguar</i>	Becker MD	Around age 5.	29	Parent not interviewed.
<i>Lamborghini</i>	DMD	Around age 5.	29	Parent not interviewed.
<i>Son not interviewed</i>	DMD	Diagnosed in the first year but precise LLTC still remains uncertain.	16	<i>Goldie</i> (mother of a son with DMD) interviewed.
<i>Daughter not interviewed</i>	Rare LLTC	Around 2 years, LLTC identified at 15 years.	20	<i>Daisy</i> (mother of a daughter with rare LLTC) interviewed.

Table 4: Participants with a known diagnosis of a LLTC at birth or in early childhood

Austin and his mother *Pear* both recounted how they received the LLTC diagnosis. *Austin* was first diagnosed with juvenile pilocytic astrocytoma (JPA), a rare childhood brain tumour, when he was two years old. In most cases, this type of tumour is benign, slow growing and does not usually spread to surrounding brain tissue. The incidence rate is estimated as 14 new cases per million in children younger than 15 years of age (Rare Diseases UK, 2018):

“I was very young when I had my first [brain] tumour, around two to three years, and I really didn’t understand what the doctors were talking about, what it [the LLTC] meant at that stage.” (Austin, YAM)

Pear separately recounted how she had received the news:

“Well when Austin was two, that’s when he had his first operation. It started with a hydrocephalus, you know, water on the brain and they sent us to see a doctor at Balchester Hospital [fictitious name] and he immediately saw that Austin had hydrocephalus, and so we then got referred to Brookway Hospital [fictitious name] where they did a scan, and they did the shunt to reduce the hydrocephalus. But then it was a brain tumour, so then he had an operation a week later, after they put the shunt in.” (Pear, FM)

Pear then explained that the tumour was benign and acknowledged that *Austin* might not have ‘survived’ had he been born twenty years earlier:

“So they took away as much as they could, and then because of the type of tumour, it was benign; they [doctors] said it is very unlikely to grow again. And I am sure in most cases that’s true. So I mean if it had been 20 years before, he

would not have survived, you know – things have progressed so quickly, so we were very grateful to the staff, not realising then that this would still be an ongoing problem for Austin.” (Pear, FM)

I then asked Pear how Austin’s condition had presented:

“Well Austin couldn’t tell me when he was little. He did all the normal things as a baby does, but then this stopped. He would cry lots if I left the room, he would be so upset, and friends, other people said ‘Well you know he is a boy, they are very clingy to their mums’.” (Pear, FM)

Pear also described when ‘things just stopped’ and because of this ‘concern’, she took Austin to see their GP because:

“Austin wasn’t eating properly; there were all sorts of little things. He hated the washing machine being on, he would get really, really upset, so I think that it was the noise, and he only felt safe if I was there. I guess the whole world sort of spun around or was horrible, but if I was there he felt safer and if I left him, just went out of the room you know, he was just lost. I am assuming that’s what it was because he couldn’t explain it. I went to the GP and she was very good. I was so concerned about Austin. But unfortunately Austin was the exception, his tumour came back.” (Pear, MF)

Pear then spoke of her concerns about the tumour recurring:

“I mean Austin is already aware because roughly it is every ten years it [the tumour] grows. The thing is Austin is convinced that it was happening again, so I guess he could feel that something wasn’t right.” (Pear, MF)

Pear also explained how she tried not to ‘create panic’ about Austin’s recurring tumours:

“I wouldn’t say to him ‘I think that is starting all over again’ because that just makes things worse, he has got enough to think about, so I would always calm things down and say ‘I am sure there is a perfectly logical reason why this is happening’ or ‘well let’s see how you go, let’s see what it’s like this afternoon, let’s see if it has got any better or if it has got worse’ because you can’t feed a fear , because you just create panic.” (Pear, FM)

Vincent, who had cystic fibrosis (CF), a life-limiting, chronic, genetic disease, which affects around 10,000 children and young adults in the UK and usually presents in early childhood (Cystic Fibrosis UK, 2018), explained to me when he received his diagnosis:

*“I remember I was around two and a half [years] and things weren’t right.”
(Vincent, YAM)*

Vincent relied on his mother (Topaz) to explain some of the early information he had received about his LLTC:

*“As the story goes, again you would have to ask my mum, but I was crying, and a nurse apparently kissed me on the head and then tasted that I had a lot of salt in my sweat as she kissed me. And then she said I think you should ask for a salt test. So then my mum did and that’s where it moved on, so I was thankful to that nurse because before then they just thought I had all different kinds of baby problems basically until I was two and a half. Instead I had cystic fibrosis.”
(Vincent, YAM)*

Vincent recognised that he also relied on ‘his mum’ Topaz and care practitioners for support, explanations and advice about his cystic fibrosis throughout his life, but acknowledged that he was never excluded from discussions about his condition with doctors:

“Yeah, I was two and a half. They [the doctors] said I had cystic fibrosis and in the end I went to paediatrics. I had a diagnosis and staff and my mum helped throughout my childhood and continue to do so. I was in the same room with the doctors and my mum and they talked about my condition. A lot of the information went over my head when I was young but at least I was not excluded from those discussions.” (Vincent, YAM)

Topaz separately described Vincent’s CF to me, which concurred with Vincent’s explanation:

“Poorly right from the very beginning of his life. Vincent was two and a half. He was six weeks premature; he went undiagnosed for two and a half years [cystic fibrosis]. It was only when the nurse licked Vincent’s skin and tasted salt that we both suspected cystic fibrosis.” (Topaz, FM)

Topaz also acknowledged that Vincent was always included in clinical discussions about his cystic fibrosis:

“We share everything about Vincent’s condition with him. Our family are very open and honest with him [Vincent] all the time. And yes, wherever possible he [Vincent] has been part of discussions with the doctors and care staff at every stage of life. Of course, it was hard for him to take on board everything when he was two and a half.” (Topaz, FM)

Topaz then described how she provided information both incrementally and appropriately according to Vincent's chronological, physical and emotional development:

"Although Vincent and I have always been open about his cystic fibrosis since he was diagnosed at two, over the years, and as he grew up, I tried to explain his condition gradually and appropriately to him and what it holds for the future, taking on board his age, stage of life and how he could absorb such information emotionally." (Topaz, MF)

Topaz described a 'transition' point for her as a parent when she was responsible for Vincent's care and treatment, but now recognised that Vincent was an adult and living with his fiancée, Smidge.

"Whilst Vincent was young, and still to some extent now, I was responsible and had control of his health. But I then lost some of this by the changing circumstances in Vincent's health, his age, desire for independence and now his relationship with his girlfriend Smidge." (Topaz, FM)

Mini had Duchenne muscular dystrophy (DMD). As described in Appendix I, this is a progressive, genetic, neuromuscular LLTC occurring in males in about one in every 3,500 UK births (Muscular Dystrophy Organisation, 2018). Mini described to me when and how he learned about his LLTC:

"I was about five years old, I think. I never expected not to be an adult. I learnt about DMD from the internet.... I talked to my natural mum... and my shared life parents about my concerns and fears about the condition... and also subscribed to Target magazine for information [published by Action Duchenne]." (Mini, YAM)

The first part of *Mini's* quote is important. It demonstrated a fracture in his anticipated life course. Non-disabled children assume that one day they will become an adult. A LLTC diagnosis questions that taken-for-granted assumption. The above quote not only highlighted *Mini's* active involvement in seeking information about his condition, but his personal desire to overcome his own 'concerns and fears' about his LLTC.

This first section has highlighted the different mechanisms for receiving the initial diagnosis of a LLTC, either via the parents, clinicians, charities or the internet, and when and where this took place. Previously many young people with LLTCs died during childhood (Beresford, 2004). Now that life expectancy has increased in this population, it is important, wherever possible, that information about specific LLTC(s) is communicated in life to both parents and young people as early as possible.

4.3 Those diagnosed in later childhood or adolescence

Adolescence is a rite of passage that is both a biological reality and sociologically constructed as a period of time that has particular significance in different social contexts (Sharma and Cockerill, 2014). It is usually a period of rapid physical development, brain growth, emotional maturity and social development for most (able-bodied) people. However, this is not always the case for people with LLTCs, as noted in Chapter 2. Adolescence usually lays the foundation for subsequent learning and development, including learning about sex(uality).

Later childhood, for the purpose of this research, refers to the period between eight and 13 years, and adolescence between 14 and 18 years (The Open University Sexuality Alliance, 2016; Sexual Respect Toolkit, 2018). Five young adults (*Diamond, Lily, Jane, Maserati and Morris, YAFs*) and two parents (*Liliana and Goldie, MFs*) described their experiences of receiving the LLTC diagnosis during this (transition) period, see Table 5.

Young Adult (Pseudonym)	Diagnosis	YA age at diagnosis	YA age at interview	Parent and research involvement
<i>Jane</i>	Rare LLC, diabetes mellitus, and visual impairment	Informed about Diabetes Mellitus, age 6. Rare LLC diagnosed age 16 years.	20	<i>Dee</i> (Boyfriend/partner, age 52) interviewed at <i>Jane</i> 's request. Her Parents – not interviewed.
<i>Son with DMD not interviewed</i>	DMD	Son diagnosed age 8 years.	Son 16 years at time of Mother's interview	<i>Goldie</i> (Mother of two sons with DMD; One deceased child with DMD, the second age 16, and diagnosed at circa 8 years.) Interviewed.
<i>Diamond</i>	Childhood Leukaemia	9 years, but the word "cancer" was not used with her until age 15 years.	39	<i>Diamond</i> was also interviewed both as a parent and a nurse. Her own parents and husband were not interviewed.
<i>Lily</i>	Rare connective tissue disorder.	Around 13 years.	20	<i>Liliana</i> (<i>Lily</i> 's mother) interviewed at <i>Lily</i> 's request.
<i>Maserati</i>	Malignant Brain Tumour.	17, but unwell for previous three years.	23	Parent not interviewed.
<i>Morris</i>	Rare LLC.	Nearly 17 years.	24	Parent not interviewed.

Table 5: Participants who knew, or were told, their LLTC diagnosis during adolescence.

Their accounts ranged from descriptions of who received the diagnosis first, the young adult or their parent, how and from whom (from clinicians or a third party), and where they received the information (via a journal, the internet or face-to-face).

Diamond had acute myeloid leukaemia (AML) during her childhood, see Appendix I. This is a blood cancer that starts from young white blood cells in the bone marrow and occurs in both adults and children (Cancer UK, 2018). *Diamond* explained how and when she felt her childhood had stopped on account of her condition:

"I would really say that my childhood stopped by age nine. That was it. I had a brilliant childhood up until then and then that was pretty much it for being a child as such, because I knew I had something wrong with me [confirmation of her LLTC, albeit the term cancer was not used until she was older]." (Diamond, YAF)

Diamond then described her illness experience in the context of her own and others' perceptions of her potential death:

"I do remember thinking that I'm going to die because I felt so awful but it's strange really, I don't actually think subconsciously that I ever thought I would. It never entered my head that I was actually going to die really. Because I do remember my parents being in supportive groups with people whose children had died and meeting up. I'd never said [to my parents] 'Where's so and so?' So then, when we met and mum had said, 'Well I didn't know how to tell you, but they've died and do you know what some people thought you had died too'. I didn't know I'd got cancer. And of course, parents tell you that you're ill but they don't necessarily give you all the information. It wasn't until I was 15 that I knew that I'd had cancer and this was deeply distressing for me." (Diamond, YAF)

Diamond illustrates how her parents had provided some, but not all, of the full details of her illness, a feature highlighted in other research related to people with LLTCs (Aldridge et al. 2017).

For *Diamond*, waiting until she was 15 to receive confirmation of her diagnosis was particularly disruptive:

"I was a pretty confident person until I received the diagnosis, then my life changed." (*Diamond*, YAF)

For *Lily*, who had a rare LLTC, learning about and living with the increasing symptoms of her LLTC and what the future held was life changing:

"I was 12 or 13 when I first started noticing real problems. I had some problems from birth but nothing that we kind of thought were significant overall, we didn't link them together. So it was just when I was about 12 or 13 my joints started to dislocate and got very weak so I had physiotherapy. And then when I was 14 I became disabled, but I was only finally diagnosed when I was 15 [PAUSE] and my whole life changed and my future was uncertain." (*Lily*, YAF)

Earlier in her interview, *Lily* had described her hopes and aspirations to me about her future:

"Yeah, I used to go horse riding every week and was hoping to study medicine at university or be a vet. But all of that has been shattered." (*Lily*, YAF)

Here *Lily*'s use of 'shattered' suggested how *Lily*'s previous hopes and ambitions were now destroyed by her LLTC diagnosis (*Lily*, YAF).

Maserati had a brain tumour which occurs in one in four children with cancer (Cancer Research UK, 2018). His diagnosis and treatment were exacerbated by the 'initial' confusion in explaining his diagnosis to him (*Maserati, YAM*).

Further on in his interview, *Maserati* stated that he felt 'cast adrift' against a backdrop of general uncertainty about his health and prognosis, whilst trying to study and plan for his future:

"I felt adrift. I had surgery to remove the tumour when I was 18... They [the surgeons] removed 80% of the tumour. This was followed by radiotherapy... Because of their incompetence... the hospital did not know it was cancer. Even when they knew it was cancer, they thought I had a prostate cancer. A bit different, heh? [Some missing words]. I had what the doctors called a peanut brain cancer, the size of a grapefruit which at the time was the size of half my head... they also thought I had migraines caused by A Level stress. I wanted to go to university." (*Maserati, YAM*)

Similar to *Diamond*, even when the young adult knew or sensed their diagnosis, parents did not always wish to discuss it with them. *Jane* had a rare life-limiting condition which, because of the small numbers with the condition, I do not wish to name here for risk of identifying her. *Jane* was also diabetic and partially sighted. Although *Jane's* parents seemingly were trying to protect her, *Jane* told me:

"If they loved [Jane] enough, they should have explained her diagnosis to her [sooner]." (*Jane, YAF*)

It transpired that *Jane* was only told about her LLTC when she was 16, although her parents had known about her condition for some time. My further questioning during the interview did not elicit a response as to why her parents had not told *Jane*:

Maddie: "So you knew your diagnosis when you were 16?"

Jane: "Yes."

Maddie: "So throughout your childhood you were, you felt fine and..."

Jane: "Yes, I had diabetes and eyesight problems but it was never concluded that it was a life-limiting condition... anything by doctors."

Maddie: "Had your parents known before [you were 16] and not said anything?"

Jane: "Yes, yes, and then I knew and only heard from my cousin."

Although *Jane* was living independently of her parents, she reflected on the periods when her parents controlled most aspects of her physical care. She felt a need to take responsibility for the future, where both she and her partner, *Dee*:

"share my care and treatment and do all my treatments" (Jane, YAF).

Diamond also explained that her parents had not used the word 'cancer' when describing her condition. She described the shock of learning the details of her diagnosis when she was 15 via a radio broadcast in which both she (and her condition) were discussed, without her knowledge and prior permission:

"I'd gone for blood tests; they'd said yes, my condition is in remission. I think I remember thinking what on earth does that word mean? But obviously over time [I] realised that remission is good and that's all we need to hear really... I think it was a bit of a shock when I was 15 and told that I had cancer. Somebody, actually my mum had taken me for some holistic therapy because of all the health problems that I was having and she [the therapist] spoke about me on the radio, that she was seeing this 15-year-old young lady that had cancer and I said to my mum, 'Surely that can't be me? And Mum replied that it was'" (Diamond, YPF)

Discovering your diagnosis via a radio broadcast (*Diamond*), or as in *Jane's* case, via a cousin or the internet, rather than hearing directly from clinicians or parents, raised important questions about how information related to conditions should be effectively communicated between clinicians, young people and parents.

Maserati recounted his frustrations about the uncertainty of 'pinpointing his brain tumour' (*Maserati, YPM*) that had taken several weeks to be confirmed. Although *Maserati* recognised in his teens that he was unwell, he had spent a relatively healthy and 'normal childhood and adolescence until he was 16:

"I went to a normal school not for disabled kids.... I started getting symptoms at 16 or 17... They were so bad that I couldn't get out of bed... I was throwing up all the time... I took the strongest codeine that I could at the time... anyway finally at age 17... my mum made me see the GP who told me I had a rare tumour... which had probably been growing for a year plus... yeah... a rare tumour." (*Maserati, YPM*)

Maserati's world had been torn apart following confirmation of his brain tumour. This impacted on his self-confidence, self-esteem, his hopes and aspirations for the future, to go to university overseas; his world had been disrupted and he found himself in limbo in being unable to escape from his illness. *Maserati* was in a transition phase between completing school studies and hoping to go to university, whilst at the same time receiving invasive treatment for cancer.

Morris had a genetic life-limiting condition caused by an abnormality on the X chromosome, mostly affecting males (Rare Diseases UK, 2018). *Morris* described how his illness experience had called into question his future, whether he would live or die, and the uncertainty of his life course, highlighting another transition point between being relatively well to becoming ill:

“I was really fine until I was nearly 17 and then things changed completely for me... no one has said anything to me about my future... whether I shall die... when I shall die... as to my future....” (Morris, YAM)

Lily’s mother, *Liliana*, described how the diagnosis of Lily’s LLTC had impacted on Lily’s social and emotional development during her teens as well as on other family members:

“It’s just at a time when girls and boys they’re becoming more aware of each other, they start dating and things like that, and because she lost that connection, she became deprived in a way and I think initially there was a little bit of contact. But it’s a very difficult age, they’re all suddenly becoming young adults and their lives are changing and, they’re then going on to college, they’re all forming different relationships and kind of [Lily] was left behind.” (Liliana, FM)

Liliana’s discussion about her daughter’s future highlighted how Lily’s condition made her different from her peers and brought into question how Lily’s life would pan out in future. [This was a concern also reflected by Karlsson et al. (2014), in research related to uncertainty in adults with LLTCS].

Liliana then talked about the uncertainty of Lily’s diagnosis, which meant that her ‘future remained unclear’ and created a particular transition point for both Lily and *Liliana*:

“Yes, she was 14. She was receiving some physiotherapy. They thought there was a possibility there could’ve been some sort of rheumatoid arthritis or something like that, but then that was ruled out and the future and her diagnosis was and remains unclear.” (Liliana, MF)

Goldie (FM) described the “terrifying and clumsy patterns and lack of mobility when Toyota was five” (her 16-year-old son with DMD, whom I did not interview).

Goldie had suspected Toyota “was not right” from early childhood:

“Toyota was nearly eight. He would get up off the floor like Bambi... he would climb stairs like Bambi... you know the cartoon character? For me, he was a textbook case of Duchenne’s, visually, mobility, coordination, everything. Various health visitors missed his diagnosis on developmental screening assessments during his childhood. It was me that questioned the diagnosis. I raised my concerns about Toyota’s condition at his school at the age of five and I was more or less told by school that I was a paranoid mum and to stop fussing. It was only during my child care course, I mentioned the signs and symptoms to the programme trainer and she advised me to see a paediatrician immediately, so I did and then at last, I got a diagnosis.” (Goldie, FM)

The findings in this section have highlighted that participants who received their diagnosis or confirmation later in life were likely to face greater disruption, particularly if they had grown up “*thinking I was normal*” (Lily, YA) and “*I was fine and pretty healthy until that moment*” (Maserati, YA). Participants did not directly acknowledge their reduced life expectancy, but the stories presented were those of truncated trajectories and individual concerns about what the future held. For young adults with LLTCs, adolescence may be a particular time of severe upheaval, instability and disruption as hopes for a ‘normal’ and ‘healthy’ emerging adulthood may change or be interrupted because of their LLTCs. The LLTC may impact on many aspects of their lives, such as self-image and self-esteem and uncertainty about the future (Abbott and Carpenter, 2016, 2012).

4.4 *Those with a recurring life-limiting and/or life-threatening condition(s)*

As stated in Chapter 2, there is an emerging literature about the long-term implications for young adults who have survived treatment for a childhood, life-threatening condition, such as childhood leukaemia, and about the risks of its recurrence (Trusson, Price and Prior, 2016; Balmer, Griffiths and Dunn, 2015). This may create another uncertain transition phase for young adults and parents.

Four young adults, *Austin*, *Maserati*, *Morris* and *Diamond*, and one mother, *Pear*, expressed their fears and concerns about the recurrence of the LLTC. This section highlights that the recurrence or threat of recurrence of the LLTC ‘casts a shadow’ over participants’ lives, thus creating disruption which may present deep uncertainties as to what the future may hold for both the young adults and their families (drawing on my model of the life course, see Figure 2, Chapter 2). Maintaining a positive attitude was regarded as an important strategy by young adults and parents in addressing uncertainty and decline, albeit that they continued to live with the reminder and uncertainty of whether a previously treated condition might one day return (*Diamond*, *YAF*, *Morris*, *Austin* and *Maserati*, *YAMs*, and *Pear*, *MF*). The issue I now explore is the flux between potential disruption and the uncertainty in their lives.

Austin, as I explained in Section 4.2, was first diagnosed with a benign brain tumour when he was two years old. The tumour was surgically removed and both *Austin* and his mother, *Pear*, were advised that the tumour would not recur. When he was 13, the tumour recurred. *Austin* described its recurrence:

“I had that diagnosed when I was two, we thought that was it. Then it grew again [the tumour] at 13. The doctors I saw in Brookway Hospital [fictional name] thought this as well... and that the tumour would not recur, but it did

recur, and again when I was 28. I'm afraid it will recur again. It seems to re-appear every decade or so and I am very worried about the future." (Austin, YAM)

Pear separately detailed the history of Austin's recurring brain tumours:

"Yes, it re-grew when Austin was 13. That was strange. Let me explain what happened. Austin was at school, he did get headaches and he could tell me about them, he would come home from school, he would have a bad headache because obviously certain situations at school made it worse, echoed assembly halls, really bright lights or you know, just children shouting, and he became much more unstable on his legs, and he came home from school and he was dragging his legs, he said 'I had to drag my leg along because I can't lift it up properly' and his left eye was watering, so we took him up to Brookway again and we didn't leave the hospital until he had had a CT scan [see Appendix A], and then it became apparent that the tumour had re-grown and Austin had further surgery. Once again, we were advised by doctors that it should not re-grow." (Pear, FM)

When I asked Pear if the tumour had grown again after Austin was 13, she described in detail:

"Well Austin finished school, did his A Levels, went off to University and got his degree, came back home, did some voluntary work at his old secondary school and went in there every day. He had a little timetable that he followed, and he came home one day in January and he was very tired, and he said 'I'm going to go to bed early because the keyboard on my computer keeps jumping around', so I said 'Okay, yes you have had a hard day, you get to bed early, that's the best thing' and when he woke up the next morning he couldn't lift his head up from the pillow and he was curled up in a fetal position, he was lying head down, his

legs were underneath him. And Austin's fists were clenched, he was completely tight. And I had to rub his fingers to ease his pain. The pain must have been excruciating and he said 'I feel my head is going to explode' and I said 'It's all right, come on you will be fine, you are very tired, move your fingers a little bit. I will go and make you a cup of tea and we will see how you go'. I managed to sit him up, he had a cup of tea and was promptly sick, so I phoned the GP and they were brilliant, the GP came round and gave him some anti-sickness tablets which cleared up the sickness and he said, 'It looks like it could probably be what you say, maybe a bug, but I am a little bit concerned. If he is no better by this time tomorrow, you ring for an ambulance straight away, don't hesitate'. Well although his sickness had stopped, he really wasn't any better so I did phone for an ambulance, and so then they came and they took him up to Barchester Hospital and then from there they did a CAT scan, and then they sent him up by ambulance back to Brookway Hospital and there was a blood clot and the tumour had regrown and he had yet another operation." (Pear, FM)

I then asked Pear how she felt about Austin's risk of getting another tumour:

"But as it turned out, the blood clot had been caused because the tumour had regrown, and they had stopped the scans you see when he was 28, they stopped the scans because he had been fine since he was 13, but they [the radiologists] are now going to do them every year. We worry about the future, whether the tumour will grow again." (Pear, FM)

The harrowing descriptions of Austin's recurring brain tumours highlighted his transitions from wellness to illness, the disruption Austin experienced in his education and hospital treatment interventions.

Pear broke down in tears at this point and we both elected to stop this interview with an option that she could contact me again in future if she wished to. I also explained that she might wish to explore these issues with a trained counsellor, given the circumstances of *Austin's* condition.

Maserati, who had recently stopped treatment, explained that:

"After a lot of messing about, the brain tumour was eventually diagnosed and I stopped treatment six months ago... for the time being that is, until they [the doctors] know if the tumour has recurred again, and this worries me. I am concerned it will return." (*Maserati, YAM*)

Diamond described 'health problems' which occurred when she was 14, leading her to suspect a recurrence of her leukaemia as her symptoms felt 'similar':

"I was about 14, we had lost touch with the hospital for some reason, but I was having lots of health problems during that time. I was always picking up every cold, every sickness bug, everything really. I was feeling very tired again. I thought the [cancer] was returning again. It felt similar." (*Diamond, YAF*)

Diamond's fears of recurrence were not isolated occurrences:

"It's a difficult time through adolescence with hormone changes. But I had lots of headaches and stomach-aches and symptoms so, after going back to the GP, she said 'You must be getting follow-ups still?' and my mum said 'Well, no. For some reason it's all stopped.' I really thought the pains and aches meant the cancer was returning." (*Diamond, YAF*)

Diamond also described her fears of a cancer recurrence when she was planning to have children, as well as the impact of her previous treatment with respect to becoming a parent:

“I had polycystic ovaries [see Appendix A] and I was unlikely to be able to conceive without fertility treatment. I was seen by a couple of SHOs [Senior House Officers] before being sent home. They told me that I would be unable to conceive because of my childhood cancer treatment. I really wanted children but I have always feared the cancer would return.” (Diamond, YAF)

The findings in this section have highlighted the fears that the recurrence of a brain tumor or blood cancer such as leukaemia create in young adults with a LLTC, creating deep uncertainties for the young adult, particularly in relation to educational opportunities (*Maserati YAM*) and reproduction (*Diamond, YAF*).

4.5 Those with an unnamed or rare life-limiting and/or life-threatening condition

Some LLTCs are so rare that there is no name or label for the specific condition (Rare Diseases UK, 2018). There are benefits in having a named LLTC: it provides a point of reference and therefore some degree of certainty for the young adults and parents; it helps both they and the young adult anticipate what may happen next and allows preparation and planning. It also may enable people to access specific support or specialist services (Fraser, 2013). Each year, approximately 6,000 children are born in the UK with a rare LLTC, often without a name (Rare Diseases UK, 2018). The reasons for the lack of diagnosis or a name may range from its rarity, to an unusual presentation of a known condition, to unnamed genetic anomalies. Without a confirmed diagnosis, clinicians may struggle to appropriately advise parents how their child’s illness will affect them, how it will progress, and how they should plan for the

future (Fraser et al., 2014). Added to this uncertainty is the creation of being a ‘one-off’, with the risk of being marginalised completely as an individual or a family.

This section addresses whether the LLTC was unconfirmed or had not been given a label. I address the findings from young adults and parents who were uncertain as to when a diagnosis was made or whether the LLTC was confirmed. Three participants (*Daisy, FM, and Jane, YA, and Liliana, FM*) were living with a condition without a label (*Jane YAF*), or experienced a diagnostic label that was not identified until later in life, or where the label or diagnosis was being re-considered (*Lily YAF*).

In some of the rarer LLTCs, diagnosis may not be confirmed, or it may change, or the condition may not be recognisable until later in life, if at all (Fraser et al., 2014; Rare Diseases UK, 2018) as *Lily* described:

“I was already 14, and they don’t take children who are already 14 [at that children’s hospital], so I was referred to Topway Hospital. I had to see the consultant privately, because the Newbase Hospital didn’t think that there was anything wrong with me, so basically my rare condition which was then undiagnosed, they felt it was all psychological, and they couldn’t put a name to it.” (Lily, YAF)

As well as the uncertainty around her diagnosis, *Lily* found herself between children’s and adult health care services, as well as having to initially pay to see a doctor to try and ascertain her diagnosis.

When I interviewed *Daisy (MF)*, she too had not received a confirmed diagnosis for her daughter’s LLTC. The lack of a firm label or diagnosis made it difficult for *Daisy* to plan for her daughter’s specific and specialist needs in the future, and explain her condition to her daughter, adding to the uncertainty and disruption in both their lives:

“Although we knew my daughter had a life-limiting condition in early childhood, it took from the age of 9 to the age of 12 to reach a provisional diagnosis of some form of developmental delay. But even now this isn’t fully confirmed, there is no name for what Buttercup [her daughter – not interviewed] has, and it’s hard for me to plan her future, let alone to explain her condition to her.” (Daisy, FM)

In the following, *Liliana*, *Lily*’s mother, illustrated the indeterminate nature of *Lily*’s diagnosis, reinforcing the idea that it involves a process rather than being an ‘either/or’ in-between feature of liminality:

Liliana: “When I first went to see a consultant and he explained, what was wrong [with Lily] and with the hypermobility, although she had all these gastric problems, even the hospital themselves weren’t making the connection between the two and it wasn’t until 18 months later when we went back to see the consultant and I just happened to touch on it, he asked more questions about her stomach problems, and then told us that it was all related. So again, even though she was in hospital being treated because of her stomach problems, nobody made the connection between the two it was very difficult to get a whole picture of what was wrong. Fortunately, we see a consultant about the gastric problems who picked up lots of different things, for instance the joints, the problem with the autonomic system, the picture started to come together then. That made it a lot easier; because I think prior to that we were sort of in the dark, no real diagnosis.

Maddie: She was 13 when she was given some diagnosis?

Liliana: Yes, she was 13, yes. She was receiving some physiotherapy. They thought there was a possibility there could’ve been some sort of rheumatoid

arthritis or something like that, but then that was ruled out{pause }But even now we're not sure" (Liliana, FM)

The 'sense-making' and attempting to integrate the diagnosis or emergent diagnosis with daily and family life was particularly disruptive for both *Lily* and *Liliana*, placing them in an uncertain and liminal state.

Goldie told me that she had questioned her son's health and condition for several years before receiving a confirmed diagnosis of Duchenne Muscular Dystrophy:

"I did not know my son definitely had DMD until he was eight years of age. I had my suspicions. I had him at 16 years [old]. I was incredibly young. I was probably too young. I noticed his walk was strange from about 18 months and kept mentioning this to my GP and the health visitor but they didn't seem to take much notice." (Goldie, FM)

This transition between suspicion and confirmation of the diagnosis was both abrupt and gradual for *Goldie*, who was a teenager herself when she had two children, both of whom had DMD and one of whom had died. *Goldie* had suspected, and eventually received confirmation, that her second son had duchenne muscular dystrophy (DMD):

"I was living in a state of limbo and it was terrifying. Not putting a name to it was frustrating." (Goldie, FM)

Two young adults stated that they were unsure when their DMD was confirmed (*Alfa Romeo* and *Fiat*, YAS). *Fiat's* parents were not interviewed at his request, so I was unable to ascertain when his DMD was diagnosed. *Fiat* explained that his brother had died of DMD at the age of 16. *Precious*, *Alfa Romeo's* mother said that:

“Alfa Romeo started to walk awkwardly when he was about three or four years, I think, and he was then investigated for ‘Duchenne’s’. But until then we had no idea what was wrong with him.” (Precious, MF)

The findings in this section suggest that the absence, delay or lack of a label or diagnosis created deep uncertainty and disruption in the lives of both young adults and parents, some of whom already felt that they were living on ‘borrowed time’ (*Vincent, Maserati and Lily, YAs, and Topaz and Liliana, PFs*). Both the young adults and parents were searching for answers, often from clinicians who couldn’t necessarily provide them, in order to plan accordingly the specific requirements amidst an uncertain future. The absence of a diagnosis was viewed both positively and negatively in this study. The ‘moment’ when a name was attributed to the young person’s condition appeared to offer ‘hope and optimism’ (*Lily and Liliana*), giving people the opportunity to speak the same language about their LLTC, opportunities to create a peer group, and potentially open a new world for young adults and their families to share their ‘lived experiences’ (*Lily, YAF and Liliana, MF*) of the LLTC. The absence of a name also created frustration, hindered planning and added to the uncertainty about the future life course.

4.6 Discussion

The young adults who are central to this research are set apart from non-disabled young adults. Their experiences are different by virtue of their symptoms and treatments for their LLTCs, the conditions themselves, and through their dependency upon others, which the symptoms and treatments impose. These factors may adversely affect their quality of life and their hopes, ambitions and expectations, both now and in the future. I argue that this was essential and necessary in order to understand participants’ responses to the specific research questions about sex(uality), outlined in Section 3.2.

In this first data chapter, I have presented findings from the disruption and uncertainty which may arise in young adults' and parents' lives through four transition points: when they first received the diagnosis of a LLTC (or not); young adults diagnosed with or suspected of having a LLTC at birth or in early childhood; late(r) childhood or adolescence or those with a recurring LLTC; and finally, for those with an unnamed or a rare LLTC. Although the number of young adult participants in this research was relatively small, the findings in this chapter have wide applicability. When and how the moment of diagnosis is encountered and how the diagnosis is communicated have an impact upon the young adult's subsequent understanding and management of both their LLTC and their sex(uality). The absence of this information was deeply distressing to *Lily (YAF)*, *Austin (YAM)*, *Liliana (PF)* and *Pear (PF)*.

Timing of the diagnosis

First, I question whether having a labelled diagnosis makes a difference as to how the young adult or parent responded to and, over time, managed the LLTC. I suggest that chronologisation plays an increasingly important role in the life course and the life events of people with LLTCs and their supporters. Thus, receiving information about a LLTC may be tied to an individual age. I argue, if the young person knows they have a LLTC in early childhood, this may or may not enable that individual to address their sexuality more easily, (*Mini, Jaguar and Lamborghini, YAs*), as an emerging adult and this is discussed further in Chapter 6.

For the adolescent who has lived as a non-disabled person until their teens and is then confronted with the presence of a LLTC, *Lily, Maserati and Morris (YAs)*, changing status from a non-disabled person to a person with a LLTC may impact on their emerging adulthood and their sexuality, and these points are developed in Chapter 6.

The transition experienced by the young adults and parents sometimes involved changed social roles and locked them into a 'betwixt and between' state of life, both before and after the LLTC was confirmed and identified, or where the LLTC was confirmed and *not* named. The rights of passage framework outlined in Chapter 2 described a tripartite structure. These features include:

'separation, requiring separation from a previous status, threshold or limen, where the individual was no longer part of their original state but not yet integrated into their new state and incorporation, and where the person re-joined society having assumed a new social status.' (Jordan, Price and Prior,, 2015, p. 840)

This particularly related to the transition of receiving the diagnosis of a LLTC, whenever this occurred, and the young person's and parents' separation from aspirations that may no longer be possible or achievable within their lives, and adapting or refocusing their lives within the confines of the LLTC, such as, *Lily*, *YAF*, and *Liliana*, *MF*.

What's in the name?

As stated in Chapter 2, for most people, life is expected to progress in a timely, staged and linear fashion (Earle and Letherby, 2007; Komaromy et al, 2007). People's lives, particularly those with LLTCs, do not always follow linear and precise routes. Instead, they may be subject to abrupt changes or transition points, where uncertainty may be magnified, particularly when a person with a LLTC does not know the exact nature and name of their condition (*Lily*, *YAF*). This not only raises important issues about being unable to plan for the future, but also being unable to manage a LLTC until the diagnosis is confirmed. The absence of a diagnosis can be a particularly destabilising period for both young adults and parents.

As stated earlier in the chapter, transitions are entry points for new periods or roles within the trajectories (Hagestad, 2003). Transition includes moving from adolescence to adulthood within the illness trajectory (Elder and Johnson, 2003) as well as from wellness to illness (*Maserati, Lily and Diamond, YAs*) or from severe illness to moderate illness (*Austin and Morris, YAs*) or to sometimes feeling better, or from life to death (*Jaguar, YAM*).

Fears of recurrence

The fears of recurrence and how this disrupted lives and created uncertainty for young adults and parents were significant (*Diamond and Austin, YAs*, and *Pear, MF*). The recurrence on three separate occasions of a brain tumour was devastating for both *Austin, YAM* and his mother, *Pearl*. Following each recurrence, the surgeons had reassured both *Austin* and his parents that the tumour would not return.

For *Diamond, YAF*, the fear of recurrence was manifested in her thinking that her children would 'probably' have cancer. Her own fears were extended to whether her childhood cancer would impact on her fertility, which I explore in Chapter 7. Trusson, Pilnick, and Roy, (2016), also explored the fears women expressed about the recurrence of breast cancer.

Whether recurrence was predicted or a surprise, it created ongoing anxiety for *Austin*, instead of being free of his condition, he still felt that he was living under the shadow of his brain tumour, 'betwixt and between' (Turner, 1969, p.95), in a liminal state where *Austin* had lost his sense of being in the real world.

All these phases were interconnected and sometimes presented the unexpected (Earle, 2007), as with the recurrence of *Austin's* benign tumour, and for *Maserati*, who mourned the physical changes in his body "*loss of hair and weight loss*", his diminished

optimism and “*aspirations for his future*”. Bury, 1982, described three aspects of disruption as the chronic illness unfolded. These were:

‘Taken for granted assumptions and behaviours; breaching of commonsense boundaries and recognising the importance of someone needing help as their body deteriorates.’ (Bury, 1982, p. 171)

Bury’s (1982) concept of illness as a ‘biographical disruption’ arose from his research with 30 people with rheumatoid arthritis. This was an important study addressing their illness experiences. Bury argued that chronic illness disrupted ‘normality’ and began the process of the individual’s reassessment and focus about their future, including their hopes, fears, aspirations and the practicalities of managing their daily lives. So for (Diamond and Austin, YAs, and Pearl, MF) recollections of intensive treatments served as reminders of the disruption that were created in their lives and were likely to continue.

The uncertainty of the life course has, and will have, a lasting impact on young people’s lives, particularly the fear of recurrence of a once treated LLTC, such as cancer or a brain tumour, only to witness its unwelcome return, not just once but possibly at several periods during a young adult’s life (Austin, YAM).

The findings in the previous three sections have revealed the theme of existential (living with and dealing with) uncertainty, in particular, how these related to when, how and if the diagnosis was confirmed: ‘the pivotal moments’ of how the information was received, and implications for the young participants’ future (Lily, Austin and Diamond, YAs, and Liliana, MF). These moments, transition points, were sometimes accompanied by the individual’s ‘transition’ to a ‘sick role’ (Parsons, 1951) and expected compliance with clinicians and medical guidance.

4.7 Conclusion

This first data chapter was necessary for the readers' understanding and to lay the foundation about the individual participants in this research and its significance to the participants' sex(uality). Individuals living with an LLTC often have different and sometimes additional barriers to discovering and enjoying their sexual rights. A certain level of knowledge about the participants' condition(s), symptoms, diagnosis, and treatment of the LLTCs is a precondition for critical engagement with, as well as appreciation, of the findings in the following chapters. I argue that unless the reader understands the world of a young adult living with a LLTC, they cannot fully comprehend the young adults' specific needs about sex(uality).

The uncertainty around the young adult's diagnosis of their LLTC was an important and compelling theme in this first data chapter. Thus, this first contextual data chapter has merited separate attention in relation to the impact and pivotal moments related to a diagnosis and how this affects young adults' and family lives present and future lives. Uncertainty in relation to an unknown life course may affect the young person at different transition points, depending on when and how the young person receives their diagnosis. How the uncertainty and pivotal moments of diagnosis of the LLTC link to the importance of relationship and sex education (SRE) is the focus of the next chapter.

Chapter 5 Sex and relationship education

Introduction

Sex and relationship education (SRE), and the rights to intimacy and sexual relationships, are interlinked (Liddiard, 2011; Blackburn, 2002). This chapter is divided into four sections. I begin by setting out the distinction between ‘formal’ and ‘informal’ SRE and what this meant to the young adults with LLTCs in this research. I go on to discuss the ‘formal’ SRE these young adults received; and where, when, and whether this was appropriate to the participants’ individual needs. Then, I explore ‘informal’ forms of SRE and how the participants accessed these and their personal preferences. Finally, I explore parents’ considerations about ‘formal’ or ‘informal’ SRE and the provision or absence of ‘specialist’ training or specialist SRE related to a young adult’s specific LLTC, and whether this was helpful and important. This section also looks at the availability of specialist education for supporters, such as partners, parents and care practitioners.

Throughout the chapter, I consider whether the presence or absence of ‘formal’ or ‘informal’ SRE influenced how young adults with LLTCs were able to engage in social, sexual and intimate relationships (see Chapter 6), and may have contributed to life course disruption. At the end of the chapter, I draw the data together and provide some early conclusions, which are explored further in Chapter 8. As in the other data chapters (Chapters 4, 6 and 7), I adopt a life course perspective to my analysis, focusing on uncertainty, liminality and biographical disruption on the findings from all four groups of participants.

5.1 *Formal and informal sex and relationship education*

As discussed in Section 2.3, both ‘formal’ and ‘informal’ sex and relationship education (SRE) have an important place for people with disabilities (see Liddiard, 2018; 2014; 2011; Frayman and Sawyer, 2014; Kelly, 2013; Abbott and Carpenter, 2012). ‘Formal’ SRE programmes may improve knowledge and awareness. Such programmes do not necessarily change behaviours or the information applied.

Differentiating between what constitutes ‘formal’ and ‘informal’ SRE was an important theme emerging from my data analysis; in particular, how young adults received SRE throughout their lives and not just at school or college. Liddiard (2011) argues that ‘formal’ sex education for disabled people mainly focuses on the biological and clinical and is too narrow (see Jackson, 1999; Corlyon and McGuire, 1999), and not on their physical and emotional needs.

In this research, ‘formal’ SRE was usually provided in schools and colleges, and sometimes in universities. ‘Informal’ SRE opportunities included talking to friends at school or college, either face-to-face or online, and accessing information via the internet and other social media. Most young adults in the research stated that they had received some information, either formal or informal, about “sex and relationships at some stage” (*Fiat, YAM*) during their lives. When, where and how this took place varied. For example, *Austin* did not receive formal SRE until he went to University:

“My formal [SRE] only happened when I went to university and was doing my degree in disability studies.” (Austin, YAM)

Several young adults (*Lily, Maserati, Morris* and *Fiat, YAs*) said that ‘formal’ SRE was a distinct subject, and usually formed part of the school curriculum:

“Sex ed. was on the school curriculum, but it was designed more for a standard healthy person, and what is expected of them, not people like me.” (Lily, YAF)

Other participants said that ‘informal’ opportunities to learn about SRE were provided in discussion groups, such as the *Sunshine* or *Terracotta hospices*’ transition groups.

Most young adults (for example, *Mini, Fiat, Lamborghini* and *Lily*, YAs) told me that they had received some formal SRE at school, college or in a hospice:

“I had formal sex education at my special school.” (Fiat, YAM)

“I received sex ed. in mainstream school but nothing specifically about my condition.” (Lily, YAF)

“Yeah, sex ed. was touched upon in my specialist college, but not a lot of formal stuff.” (Lamborghini, YAM)

“There was a lot about independence training, some formal sex education provided by health staff in the hospice.” (Mini, YAM)

Morris had attended a mainstream school in Ireland where there was no SRE:

“I was brought up in Ireland, but I had no formal sex education at school. I started out at a normal school and it was quite religious so definitely no sex ed. there.” (Morris, YAM)

Jane, who attended a special school for people with visual disabilities stated:

“Well I didn’t have anything formal set out in sex ed. at blind school.” (Jane, YAF)

In her first interview with me, *Jane* described some of the contraceptive advice that she had received at a school for visually impaired people:

“Where you put the condom or the rubber on, you know, we watched videos and, yeah, but it was never really explained to us the importance of being careful and pregnancy and all that stuff but not covered in general sex ed. at blind school.”

(Jane, YAF)

This raised questions about what young adults regarded as ‘formal’ SRE and what they regarded as ‘treatment’, and if they associated a link between the two. So for *Jane*, SRE focused specifically on her treatment aspects of contraception and prevention of pregnancy, rather than broader areas of adolescence, reproduction, pregnancy and childbirth education.

When I discussed the provision of formal SRE with parent participants, five stated that they ‘thought’ their son or daughter had received ‘some formal’ SRE at school (*Ruby, Precious, Liliana, Emerald and Goldie, MFs*):

“I think he [her son, not interviewed] had PHSE, biology, general education... and formal stuff at school and college.” (Ruby, PF)

Goldie and Precious acknowledged the importance of ‘formal’ SRE in both mainstream and special education:

“Discussing sex should not be a taboo subject. They should be formal topics for discussion. My son had formal sex education in his mainstream school, but nothing formal as yet has been provided in his current special school.” (Goldie, MF)

Four parents (*Opal, Crystal, Daisy and Precious, MFs*) felt that the provision of formal SRE at school or college supported and reduced “the burden and pressure on parents” (*Crystal, MF*) to provide SRE.

Some parents (*Goldie, Aqua Marine, Crystal, Liliana and Topaz, MFs*) recognised that this was:

“a shared responsibility between parents, teachers and care practitioners.”
(*Crystal, MF*)

Topaz said that not all care practitioners felt comfortable discussing sex but suggested that they should be prepared to:

“I think that they [hospital staff] would be embarrassed talking about sex. Well, if they are, they shouldn’t be in that job, should they?” (*Topaz, MF*)

Some parents said that they were uncertain or could not remember whether their son or daughter had received ‘formal’ SRE or where this was provided (*Precious and Pearl, MFs*):

“Not sure if sex ed. was covered at school but it would have been easier if they [the school] did this, as this was not an easy subject for us to talk about with [her son with DMD]. My husband finds talking about sex with all of our kids quite difficult and harder with our disabled son.” (*Precious, MF*)

As noted in Chapter 3, recruiting fathers to participate in this research was difficult and none took part in interviews. All the female parents stated that it was they, rather than the fathers, who mainly ‘talked about sex’ with their sons or daughters, for example *Topaz and Crystal (MFs)*:

"No, we haven't had any of that [sex ed.] at the adult clinic and it would be mainly my job and now Smidge's [Vincent's girlfriend] to discuss sex ed. matters with Vincent. My husband leaves this kind of stuff to me or Smidge, and he [her husband] has always done so. He finds it difficult to discuss this kind of thing with any of our kids, but particularly Vincent." (Topaz, MF)

Care practitioners described a varied picture about what constituted 'formal' and 'informal' SRE for young adults, as well as training for parents, partners and the care practitioners themselves:

"Altogether, there is limited formal training about sex for us [the staff]. There's some for the young people, but not much for parents who use the hospice." (Pine, CPF)

"Yes, we certainly have Parents' Groups but not really where 'formal' discussions take place about sex. [Discussions are] more about independence and how young people would manage living away from home." (Primrose, CPF)

"We have a transition group that meets regularly at the hospice and the group discusses sexual matters together. We are setting up Sexuality Champions in the hospice to facilitate more formal discussions about sex between young people and staff both in groups and on a one-to-one basis." (Pine, CPF)

In summary, all four groups of participants recognised the importance of formal SRE. Women were mainly seen as responsible for addressing SRE within the family or at home, and in either 'formal' or 'informal' settings, although the emergence of transition groups facilitated 'informal' discussions to take place within a 'formal' setting. The role of sex educator appeared to be unclear: overall it was perceived as someone else's role. Following their involvement in this research, *Sunshine Hospice*

established Sexuality Champions to help address sex education which was a welcome development. This leads to the next section about the location, timing and nature of SRE.

5.2 *Where, when and how*

The four groups of participants presented a varied picture of the sources and resources of 'formal' SRE and where this took place.

Most young adults discussed the different stages at which they had received formal SRE (or not). Their experiences varied in relation to the location, timing and how SRE was provided. Two young men, *Alfa Romeo* and *Vincent*, stated that they had been discouraged from attending 'formal' SRE programmes at school as it was felt that "the content would be irrelevant" to them, firstly because they were disabled, and secondly because they were likely to die. If they were going to die before adulthood, SRE would not be necessary.

Similar views were noted in research with young adults with spina bifida and/or hydrocephalus. Young adults were discouraged or even denied access to SRE and encouraged to attend the hydrotherapy pool instead, as this was perceived to be more beneficial than attending SRE (Blackburn, 2002).

In addition, religion was sometimes described as a barrier to both formal and informal forms of SRE:

"Wherever sex ed. was provided, formal or informal, religion always got in the way for my family. They are very religious." (Lamborghini, YAM)

Most young adults who had received SRE stated that they had received this during their teens and within mainstream schools, special schools, colleges, residential units, hospitals, or in hospices:

“I had PSHE in both mainstream and special school during my teens.” (Mini, YAM)

Maserati stated he had only received SRE “such what it was” in his mainstream, secondary school:

“I suppose I did some sex education at [mainstream] school. We were introduced to this subject, such what it was. We were shown a video at age 11 about reproduction but it didn’t cover anything I didn’t already know. Between then and PSE [his term] at 16 years... there was no real sex education at all. And then I became ill.” (Maserati, YAM)

Fiat said that he had received SRE in a residential setting. His training was optional:

“My residential care unit provided sex education in my late teens. We were invited to sex education but some [young people] refused to attend. I attended but didn’t have to.” (Fiat, YAM)

When I asked, *Fiat* did not elaborate as to why people “refused to attend” SRE but he said that “the classes were not that helpful” (*Fiat, YAM*).

Austin, Vincent and *Diamond* (YAs) reported that they had long periods of absence from school on account of treatments and hospital in-patient stays associated with their specific LLTCs. They ‘missed out’ on SRE courses and, in *Vincent’s* and *Diamond’s* case, there were no’ catch-up opportunities’:

“A bit [of sex education] when I was doing my degree in disability studies but not related to my tumour and how it would impact on relationships. Nothing at school, but I was in and out of hospital though and missed the sex ed. programmes there.” (Austin, YAM)

“Oh, no. Sex education not through school, no. A bit at the hospital clinic but I was often away from school because of my cystic fibrosis so missed out and there was no catch-up programme.” (Vincent, YAM)

Austin, Vincent and Diamond (YAs) also acknowledged that treatment programmes often interfered with their overall education, school examinations and SRE:

“Because of all the treatment, I was off for long periods and then I suffered from depression which was never picked up [by clinicians] all through my teens and suffering with school-phobia and I was very rarely at school. So, yeah, little sex ed. at school, little education altogether at school because of my condition.” (Diamond, YAF)

Diamond not only talked here about her absences from school, but addressed features arising from these; notably her depression and school-phobia. I suggest that this was a pivotal period in her life and contributed to some of the biographical disruption in her life.

Young adults with LLTCs have disrupted lives. They have frequent visits and admissions to hospital and absence from formal education, including SRE (Brown, Coad and Franklin, 2017). In addition, many adults with LLTCs have accompanying physical, sensory and/or cognitive impairments, as well as experiencing intermittent periods of acute and chronic illness, and periods of treatment (Brook, 2014). Many of the young

adults I interviewed had lived in residential settings and spent considerable time in hospitals or in hospices and were:

‘excluded from most of the dominant socialisation processes that help teach and prepare people for love, sex and intimacy.’ (Davies, 2000, p. 181)

In the past, SRE was often denied or unavailable to disabled people (Shakespeare, 1996), and for some disabled people this still remains an issue. Shakespeare, Gillespie and Davies (1996), Garbutt (2010) and Hollomotz (2010) argue that the denial of even basic anatomical knowledge of reproduction to disabled young people remains tied to dominant notions of infantilisation:

“We were told that our son would die before he was 16 years. So sex ed. would be unnecessary. Our son is very aware of death and can talk about it, but sex ed.? That’s different.” (Precious, PF)

Curriculum content and preferences

Appropriate education and training is essential for young adults, as well as those providing care for young adults with LLTCs, and those making transitions from child to adult services (Doug et al, 2011a, 2011b).

Alfa Romeo suggested that his SRE was limited to biology, suggesting that biology was separate from SRE and not seen as part of an integrated curriculum:

“I did not cover sex at school; we did the biology and emotional stuff but no sex education.” (Alfa Romeo, YAM)

Alfa Romeo and *Jane* had both received some ‘formal’ SRE, even if they did not recognise it as such, which I argue indicated some confusion about both theirs and other people’s understanding of what SRE included and whether it was analogous to their treatments. *Vincent* explained that he first received SRE from a doctor in hospital:

“I was 15 and at Faithful Hospital. I was sitting with Dr Gentle and I think he said, ‘you know about contraception?’ I think he was giving me the whole basic talk about, I don’t know, sex and contraception. But I didn’t get anything else from him [the doctor] and this would have been helpful.” (Vincent, YAM)

Vincent acknowledged that he had received contraceptive advice from his hospital specialist, but he would also have liked his specialist to offer more ‘formal’ advice about his condition (CF) and how this might impact on his sex(uality). This highlighted the important links between the pathology of cystic fibrosis, its relationship with SRE (Sawyer et al, 2014) and *Vincent*’s emerging sexuality.

Views about sex and relationship education

Several participants told me that the SRE did not necessarily meet their specific requirements (for example, *Lily* and *Jane*, YAFs, *Fiat*, *Mini* and *Alfa Romeo*, YAMs, and *Precious*, *Goldie* and *Crystal*, PFs):

“Would have been useful to have specific skills for meeting people, you know, social skills with the opposite sex. Get an overview of most things. You know about sex, friendships and relationships. It’s important to us too.” (Alfa Romeo, YAM)

Alfa Romeo spoke about the importance of young adults being able to access specialist SRE, and he also recognised the importance of developing social skills. Many young adults in this research were socially isolated, particularly after leaving school or college, and especially if they were unable to obtain employment.

Several participants stated that ‘formal’ SRE often focused on heteronormative subjects and behaviours rather than specialist subjects that might be more relevant to people with a particular LLTC:

“They seemed to think it [sex ed.] was irrelevant because of her life-limiting condition [Marmaduke’s LLTC] and was just for ‘normal’ people. How wrong they were.” (Crystal, MF)

Protecting people from harm is and should be an important aspect of formal SRE for people with LLTCs. However, this was not always the case here. Five young adults acknowledged this – *Jane, Lily, Vincent, Alfa Romeo* and *Morris (YAs)*. *Lily* pointed out that:

“We need to have sex ed. that includes discussions of risk taking and protection from harm to help inform people like me to make wise choices and to take acceptable risks without being exploited.” (Lily YAF)

One young adult female, who requested anonymity, suggested that her rape might have been avoided had she received more information at school about ‘protection from harm’ and if she had been provided with SRE ‘catch-up’ opportunities when she returned to school:

“I was raped when I was 17. So that has been a problem over the years. I didn’t tell anybody initially. People know now. It [rape, child abuse] was not something

ever addressed at school or ever covered in sex ed.” (Pseudonym not disclosed to respect confidentiality)

Similarly, *Morris* described his vulnerability and the impact and pivotal period of his first ‘sexual experience’ when he had a seizure and almost died whilst he was making love to his (then) girlfriend:

“I don’t have the courage to ask out women, come out, or go out with them. You know, cos of my illness, I think it’s because of this event, making love, in the past. It had a big impact on me. I nearly died! I collapsed. I feel scared about meeting girls and having a girlfriend now. I almost died, I was unconscious. I need information to help me build confidence again.” (Morris, YAM)

After *Morris*’s disclosure to me about this experience, with his consent, he allowed me to discuss this aspect of the interview with the hospice’s Director of Care. This was to enable *Morris* to obtain therapeutic intervention through their hospice services. It led to *Morris* receiving a referral to a trained psychosexual counsellor who could talk to him about his sex(uality) and his experience in a ‘formal’ setting. This had the effect of bringing together both formal and informal SRE opportunities for *Morris* to acquire important information to overcome his fears about ‘having sex’ with someone else in future.

Most young adults had received some formal SRE at various stages of adolescence and adulthood within different environments, but with little SRE specific to the young person’s LLTC. Regular, planned and unplanned absences from school on account of severe ill health and treatments for the LLTC meant that many young adults’ lives were disrupted (Kelly and Vougioukalou, 2017). They could not attend school or college regularly and therefore did not attend SRE courses. Furthermore, there were limited opportunities available to redress those gaps in SRE provision during adolescence.

Both the young woman's rape and *Morris'* descriptions of a seizure during love-making highlighted the vulnerability of some people with LLTCs, and underlined the need to protect these young adults from harm. I suggest that, had the young adult woman been able to access SRE during adolescence, she might have been able to protect herself from sexual assault or may have been more likely to report it.

SRE is more than preventing sexually transmitted diseases and pregnancy. I suggest that the young adult woman may have found her teenage 'rape' difficult to discuss with anyone as she felt a sense of shame, personal responsibility, and had difficulty in discussing a taboo subject at this stage of her life. Similarly, I suggest that *Morris* was afraid of seeking help to address his fears about having another girlfriend following his seizure. Abusive or potentially abusive situations are often addressed better in face-to-face situations, whilst sometimes informal learning opportunities may provide support and information.

5.3 *Informal opportunities to learn about sex(uality)*

Most of the young adults in the study stated that they had accessed information about sex and relationships via friends, the hospices' young people's transition groups, talking to therapists, or reading magazines, such as *Target* (produced by The Muscular Dystrophy Association). In addition, young adults stated that they increasingly used the internet and service user websites, such as DMD Pathfinders, Tinder, Lucy's Light Blog (set up in 2014) and other social media, such as Facebook and Twitter.

Once young adults left school or college, if they were not in employment or working in a voluntary capacity, face-to-face opportunities to 'talk about sex' were more limited. Both hospices ran transition groups where young people met 'informally' on a two-weekly basis and both provided young people with the opportunity to talk with each other and the care practitioners about sex.

As already stated in Chapter 2, talking about sex was regarded by several young adults as a ‘taboo’ or ‘awkward’ subject. It was also viewed by several participants as “an embarrassing subject” (*Fiat, Lily, Diamond, Jaguar, Lamborghini, Austin and Mini, YAs*). Not talking about it may only serve to increase the stigma and taboos that exist about sex and make people more, rather than less, vulnerable to abuse (Liddiard, 2018, 2011).

Other young adults said that they found ‘talking about sex’ with friends and carers ‘helpful’, particularly when these took place at the *Sunshine* or *Terracotta Hospices’* transition groups (*Focus Group, Sunshine Hospice: Fiat, Alfa Romeo and Lamborghini, YAMs, and Jane, YAF*).

Young adults said that it was ‘easier to talk to individual friends or carers’ of either the same or opposite sex (*Austin, YAM, Lily and Diamond, YAFs*), but others found online learning useful:

“Being able to find out through the internet helped and provided me with information, particularly when I spend so much time on my own at home.”
(*Austin, YAM*)

Whereas for *Fiat*:

“Your own experiences are the best experiences of all and talking to friends helps. Some people say they learn more about sex on the internet, such as dating. That may help.” (*Fiat, YAM*)

The majority of young adults stated that they found it difficult to discuss SRE with their parents because of the ‘generation gap’ (*Austin, YAM*), stating that they “felt embarrassed” (*Austin, YAM*) talking to them. However, media debate around the

protection of vulnerable young people sometimes prompted discussion between parents and the young adults:

"I don't often talk about relationships and sexuality with them [my parents]. Too embarrassing! I think the most we've spoken about relationships and sex and stuff is when we've seen on the news about JS [not named here] and dreadful things like that." (Austin, YAM)

Two young adults (*Vincent* and *Lily*, YAs) said that they did not find it difficult to talk to parents about SRE. *Vincent* and *Lily* both felt that the complexities of their conditions made it important to discuss SRE with them:

"Talking about sex [with parents] is and should be no more difficult than discussing sensitive treatments about bladder control or my breathing." (Vincent, YAM)

From the parents' perspective, *Precious* said talking about sex was equally difficult with both her non-disabled and disabled children:

"I am embarrassed talking about sex with our non-disabled children, so discussing sex with a young person with a LLTC is even harder." (Precious, MF)

Daisy stated that:

"Nothing's ever been offered to me [about discussing sex]. They [young people] shouldn't be denied relationships or sex education just because they have a special educational need or a disability." (Daisy, PF)

Daisy also pointed out there wasn't anyone for her to talk to about her daughter's sexuality, such as doctors or nurses:

"There was nobody really with that role. I think the only person I've come across since who would have been the right person and who gave us leaflets on breast examination for young ladies was a learning disability nurse that we came across totally by accident and she spoke about sexuality and learning disability specifically. Doctors tried to pass us on to specialist health visitors, but specialist health visitors don't have the time [to talk about sex(uality)], and all they've got time to look at, is continence and sleep and bowel problems." (Daisy, MF)

Diamond described her parents' preoccupation with her LLTC and the fact that Diamond was 'different' from other 'normal' young people. Diamond felt that her parents were 'over-protective' and discussing sex with them was 'not an option'. They related to her differently:

"Being a mum of a child that had leukaemia, she [Diamond's mother] did make me feel that I was different. So I did have some issues with them when I was growing up." (Diamond, YAF)

Some parents (Precious, Emerald, Diamond and Goldie, MFs) were professionally employed in 'caring roles' (see Table 2, Chapter 3). They stated that they recognised the importance of access to SRE, but also questioned the availability of 'safe' and 'useful' information about sex for young people with a life-limiting condition (Goldie, MF). In particular, Precious noted that:

"Zephyr [not interviewed] reads Target magazine which is for people with Duchenne's and there is sometimes something about relationships there. He has made a lot of friends with other lads who have Duchenne's at Sunshine Hospice

and I know they discuss relationships, sex, intimacy and all that stuff when they get together. Sunshine Hospice holds a regular group with the young adults to discuss transition into adult life, relationships and sex and offer advice, as necessary. But as far as I am aware, I don't think there is anything for parents. I think we, me and my husband, would have been invited, if there was such a group. My husband is sure that sexuality is an issue for Zephyr, although he doesn't really talk to us about it. It's not that we mind discussing sex or anything. But we are older, the wrong generation, this sort of stuff, he would rather discuss with his friends of the same age." (Precious, MF)

Opportunities to access specialist sex and relationship education

All four groups of participants acknowledged that specialist SRE, particularly for young adults surviving into adulthood with a LLTC, was not readily available, particularly after leaving school (*Diamond, Lily, Vincent, Jane, Morris, Fiat, Jaguar and Lamborghini, YAs*). Similarly, two partners said that there was little information available for those in a relationship with somebody with a LLTC (*Smidge and Dee, Partners*):

"I learnt about cystic fibrosis from Vincent, his mum and reading up online. Managing the specifics of his breathing difficulties when we're cuddling, I hadn't a clue initially. I've learned on the job, so to speak." (Smidge, PF)

"My first wife had a chronic condition, so although I am aware of some of the practicalities of caring for someone, Jane has a rare life-limiting condition. She also has type two diabetes and is blind and incontinent. Took me some time to get to grips with these conditions and how they might impact on our relationship." (Dee, PM)

Most parents I interviewed said that they had received “little or no” (*Goldie, MF*) information about the sex(uality) of their son or daughter. *Goldie*, for example, describes how she had to take it upon herself to find out about sex. She says:

“I had to do a palliative care module to learn anything about adolescence and sex and the specifics for my son.” (Goldie, PF)

Daisy said that she had “picked up bits” of information whilst training as an educational specialist support worker. *Diamond* subsequently trained as a nurse but she stated that her nurse training:

“...lacked information about the sexuality of people with such conditions [LLTCs].” (Diamond, CPF)

Diamond went on to add:

“I would like to see [a] much more supportive network put in place, much more information provided to children and young people with these sorts of disorders and information much more widely available, so that people aren’t left feeling unsupported and also not being educated on how things might be and thinking about healthy lifestyles really.” (Diamond, YAF)

As described in Chapters 1 and 3, the young adults had a range of LLTCs (see Appendix I). One young person was married with children and two other couples were in a ‘steady relationship’. All three couples stated that they would have benefitted from more and ‘ongoing’ information about ‘the mechanics and specifics’ of sex related to their LLTC.

Most care practitioners acknowledged that SRE was not included in their undergraduate or postgraduate medical, nursing or social work training (*Ellie, Primrose, Oak and Peach, CPS*). Two GPs and a medical student reported that they had not received specialist training about sex(uality), either at medical school or in postgraduate courses, although both GPs had attended and completed the Cardiff Specialist Paediatric Palliative Care Postgraduate Diploma during their careers:

*"I would not feel skilled, experienced or empowered in this area [sex-ed.] at all."
(Elm, CPF, GP)*

This is a gap that has been acknowledged by The Association for Paediatric Palliative Medicine and Together for Short Lives (hereafter TfSL) national research group and is further discussed in Chapter 8.

"Whether [sex ed.] would be ever a priority topic I'm not sure and I think maybe GPs aren't the people that necessarily should be being targeted. Maybe it should be family support workers, social workers, parents. Rather than the GPs necessarily because not always is it a health medical issue anyway." (Ellie, CPF, GP)

"We did a little bit of palliative care but only small amounts; we did general palliative care as part of GP training to a limited amount but no focus on young people and certainly no focus on sexuality, never, and certainly not in any way from a palliative care or an end-of-life care perspective at all." (Oak, CPF, GP)

"I've just done three years as a medical student so we did two years which was pure lectures and in that there hasn't really been any emphasis on sexuality or disability at all yet. Hopefully it will come." (Peach, Medical Student, Female)

Although some specialist palliative care training was provided for doctors, it was noteworthy that both the GPs and the medical student I interviewed felt that other health and social care practitioners were probably more suited to provide specialist SRE than they were, but they too felt that they needed more specialist training:

“I never had any sex ed. training on my nursing or social work courses. The hospice has sent me on a training day but it didn’t really cover specific issues related to LLTCs and sex Ed.” (Apple, CPF)

In summary, most young adults in this research were keen to receive SRE. Their preferences as to the timing and the nature of the information (formal or informal) varied. All groups of participants emphasised that, for young adults with LLTCs, quality of life and quality of care were crucial and that must extend to include personalised SRE as part of lifelong learning (*Lily, YPF; TH, and Liliana, PF, TH*). As SRE was difficult to communicate in relation to LLTCs, and because it could not be ‘off the shelf’ advice, it was important to signpost young adults to SRE and specific information that they required, to reduce the uncertainty in their everyday lives. It was important that access to information, and the information they received, was tailored to their individual physical and emotional requirements, in an age-appropriate way, and the focus was on their quality of life, rather than on the ever-present uncertainty of death.

5.4 Discussion

For many years, people with disabilities have fought to have their sexuality legitimised and not politicised (Liddiard, 2018; Przybylo, 2013; Smart, 2009; Shakespeare, 2000). The enforced and oppressive label of ‘asexuality’ has often been imposed on disabled people by society (Liddiard, 2018, 2011). People with disabilities have been assumed to lack sexual desire and function, and therefore access to information about sex(uality) has often been seen as unnecessary (Liddiard, 2018, 2011). As this chapter

has highlighted, relationships, body image and sexuality are considerations that are defining characteristics for many adolescents who experience 'normal' health. This may be expected to assume even greater significance during, and following, a diagnosis of a LLTC (Kelly, 2013). However, adolescence applies to an age group that, whilst sharing some common features, will also be disparate in terms of individual physical, emotional and social requirements. The rate at which sexual maturity is reached does not necessarily follow a fixed pattern, underlying the need for individual access to formal programmes (Kelly, 2013) as well as informal SRE.

Research suggests that there are challenges associated with planning for and living the life of an 'unanticipated adult'. These include not having the opportunity to learn or talk about sensitive subjects such as sex and death, and living with the physical and psychological aspects of a LLTC (Abbott and Carpenter, 2014; 2012). The findings in this research resonate with those of Abbott and Carpenter and here I discuss the most specific issues related to SRE: access to (specialist, both formal and informal) SRE for people with LLTCs, lifelong learning about sex(uality), designated sex educators, undergraduate and postgraduate training, palliative care, sex education in medicine and nursing schools and, finally, balancing the risk of sexual exploitation with making safe, informed sexual choices and decisions (De Than, 2014).

Access to formal and informal specialist education

All four groups of participants recognised the importance of formal SRE. Women were mainly seen as responsible for addressing 'sex education' within the family or at home. Although Walker notes that a few fathers share this role, the biological sex of the parents and their own experience of sex education clearly influence the sex education they provide within the family (Walker, 2001). Women are still regarded as the major caregivers and hence the main health educators in the home (Walker, 2007).

The emergence of transition groups within hospices facilitated ‘informal’ discussions to take place within a ‘formal’ setting for people with LLTCs. All four groups of participants said that formal sex education was helpful but needed to be ongoing and that its focus was often too narrow and ‘ableist’ (see Liddiard, 2018, 2011; Jackson, 1999; Corlyon and McGuire, 1997), suggesting that it was not relevant to the lived experiences of young adults with specific and individual LLTC(s).

Access to age-appropriate, condition-specific SRE was frequently absent from formal SRE programmes in schools and colleges. Doug et al (2011a) noted that successful health, social care and education programmes for young people with LLTCs should be designed to respond to the young people’s individual needs. In their review of 92 research studies addressing the needs of young people aged 13 to 24 with various LLTCs, they did not specifically consider access to SRE, albeit that most of the transition programmes they reported on related to condition-specific LLTCs (such as cystic fibrosis and cancer) within health, social care and education services.

As *Lily, Vincent and Maserati (YAs)* explained, young adults with LLTCs require access to tailored, formal SRE. I suggest that this is particularly important for people with LLTCs because they do not have an easily accessible safety net of either formal or informal SRE, as their information needs are specialist and the specific LLTCs addressed in this research are complex and different; so one size does not necessarily fit all. Kelly (2013), in his research related to young adult cancer survivors, noted that sexuality, relationships and body image are concerns that are defining characteristics for many adolescents in normal health, but are even more significant following cancer diagnosis. He argues for personalised SRE related to cancer for adolescent survivors. He also notes that the provision of specialist services or individual support at the end of cancer treatment may be one measure to ensure that peer support is provided.

Lack of, or being denied access to SRE, may lead to distress, feelings of isolation and a sense that individual needs are unimportant. This was articulated in the *Morris*, *Maserati* and *Diamond* (YAs) interviews and echoed by Liddiard (2018, 2011) and Blackburn (2002) in sexuality research with disabled people. Widespread constructions of disability continue to shape the extent to which young disabled people acquire knowledge about sex and sex-related topics, whether formally or informally: contraception, sexual health, personal relationships, emotion and reproduction (Liddiard, 2018). This impacts upon sexual development and may contribute to disabled people feeling oppressed (Shakespeare, 1996). It may sometimes be regarded as a central form of psycho-emotional disablism (Reeve, 2004), and may make some disabled people more susceptible to sexual abuse (Gillespie-Sells, Hill and Robbins, 1998).

Prolonged and regular absences from school described by young adult participants meant that it was difficult to access or 'catch up' with formal SRE programmes (*Diamond* and *Vincent*, YAs). As discussed in Chapter 2, Kelly (2013) noted that public embarrassment and taboo may be coupled with a lack of access to SRE information due to prolonged periods of hospitalisation for cancer treatment. This was expressed by *Pear* and *Emerald* (FMs). Kelly (2013) also noted that discussing SRE was difficult for parents, especially as friends and peers were usually the preferred choice for such conversations.

Currently there is no statutory, specialist sex education provision for people with physical disabilities including for those with LLTCs (The Open University Sexuality Alliance, 2016; Blackburn, 2002) in the UK. At the time of completing this thesis, a government select committee has outlined the role that SRE could play in ensuring that both non-disabled and disabled children and young people understand gender, equality, sex and consent, and placing SRE curriculum on a statutory footing, within defined parameters. A recent government select committee report (2017) for SRE

discussed elements that are mainly associated with the science curriculum, but included the importance of relational aspects for young people within the SRE curriculum. For many years, these elements have been requested and articulated by young people, parents, health professionals and other statutory and non-statutory agencies and will come into force in 2020 (DfE, 2019a; 2019b).

Death was seen as imminent and inevitable by some parents (*Pear and Emerald, MFs*). Therefore, the need for SRE was not always recognised or prioritised (Liddiard, 2018, 2011; Shakespeare, 2000). Participants, whether partners, parents or care practitioners, sometimes made (mis)assumptions about what the young adult needed or wanted, thus making it difficult and sometimes impossible to have conversations about sex (ACT and Triangle, 2009).

Lifelong learning

Whilst transition into adult life may be complicated by deteriorating physical and emotional conditions, and in some cases learning disabilities, ignoring the specific sex education requirements of people with LLTCs is no longer an option, as my research findings have indicated. The mean average age of the young adult participants at interview in this research was 26 years. Young adults with DMD in this study were 'learning' to address their sexuality as 'older', unanticipated adults (Abbott and Carpenter, 2014). As well as socially constructed barriers, young men with DMD faced other limitations due to the severity of their LLTC, and because of misjudged assumptions about the nature and value of their lives which are likely to be shorter than most non-disabled people. The complicated nature of shifting expectations across a life course made planning for an adult life with a LLTC very challenging (Abbott and Carpenter, 2014). This included the recognition of the emerging sexuality of men with DMD who wanted to access, both formally and informally, information about sex after leaving school and college.

All groups of participants in this research emphasised that for young adults with LLTCs, quality of life and quality of care were crucial and should extend to include personalised SRE as part of lifelong learning (*Lily, YAF, and Liliانا, MF*). I argue that SRE needs to signpost and engage young adults to access SRE information that they personally want and to consider when it is required, firstly to reduce the uncertainty, and secondly, to minimise the biographical disruption in their everyday lives. It was important that access to information, and the information they received, was appropriately tailored to their individual physical and emotional requirements, and the focus was on their quality of life, including their sexual lives, rather than on the ever-present uncertainty of death (Goodley, Liddiard and Runswick-Cole, 2017; Abbott and Carpenter, 2014). Kelly (2013) recognises that adolescence for people with LLTCs may be disruptive and I suggest necessitates a more open-ended approach to the delivery and accessibility of both formal and informal SRE, acknowledging that:

‘The rate at which sexual maturity is reached does not follow a fixed pattern, underlying further the need for individual assessment being at the heart of personalized cancer rehabilitation for adolescents.’ (Kelly, 2013)

The sex educator

The role of sex educator appeared to be unclear. Overall, it was perceived by participants as being adopted by various individuals. No one professional group of either health or social care practitioners appeared to have this responsibility. Care practitioners, such as *Oak, Apple* and *Ellie*, said they were not trained to take on this role, even though they were tasked with providing holistic care to people with LLTCs in their work settings. For participants, such as *Vincent*, and *Topaz*, they stated that the hospital doctor had a role. For *Goldie, MF*, it was mainstream and special schools, but there was also the importance of recognising parents, such as *Goldie* and *Topaz*, important roles as sex educators (Williams, 2001). Frayman and Sawyer (2014) have

identified a similar gap in sexual and reproductive health education responsibilities in Australia for people with cystic fibrosis.

As noted by *Pine (CPF)*, see 5.1, *Sunshine Hospice* have established Sexuality Champions to help address sex education, which was a welcome development within a hospice setting. Similarly, Kelly and Hooker (2007) note that the central philosophy of cancer education programmes for non-specialist professionals emphasises the importance of improving communication between clinicians and service users. Kelly also highlights the importance of effective involvement of service users in the engagement of cancer care education programmes, including SRE (Kelly, 2013).

People with LLTCs in this research were often socially isolated and media played an important contribution in their 'informal' learning about SRE, because engagement with friends, although preferred, was not always possible. Young adults with LLTCs have much to cope with in their lives so the provision of appropriate 'formal' or 'informal' SRE was important to enable young people, parents and care practitioners to address the practical implications of growing up and the lived experience of a LLTC, as well as the intersectionality between sex(uality) on an uncertain life course.

Sexual choices and protection from harm

The Care Act (2014), see Appendix B, in England sets out the statutory responsibility for protecting adults who may be 'vulnerable to abuse' and require protection from harm. Local Authorities in partnership with health, local and neighbouring social care services share these responsibilities. Young adults with LLTCs have the right to relationships, fun and sexual expression like anyone else, without always necessitating:

‘Wrapping [disabled people] up in cotton wool [...] (Munby, 2002; cited in De Than, 2014)

Such rights, as cited in the legal case of *Pretty v UK, 2002* by Sir James Munby, President of the Family Courts in England and Wales, should be respected, wherever possible, whenever no harm will be caused (De Than, 2014). This often makes issues about accessing information about prevention from sexual exploitation difficult and this is addressed further in Chapters 6 and Appendix B.

5.5 Conclusion

In this chapter, I have presented and discussed the findings related to both formal and informal SRE. The findings have indicated that most young people with LLTCs had reduced opportunities to access formal SRE and that, when they did receive it, it was not specific to their LLTC. Participants felt constrained by the uncertainty of end of life and what the future held. Yet being able to access information and knowledge about sex(uality) throughout life was important for these young adults, as well as for those parents, partners and carers who continued to provide support in adulthood. Care practitioners admitted that they required skills and confidence to address sex(uality) across a wide range of young adult settings in order to build confidence and trust with young people, as well as with parents, partners and other carers.

Guidance and bioethical literature increasingly advocate the participation of adolescents in decisions about their health care (Day, 2016). Yet, little sex(uality) guidance was offered to elucidate what this involvement looked like in practice, for young people, their partners and their parents in this research.

Chapter 6 The meanings and experiences of sex(uality)

Introduction

This chapter discusses the participants' meanings and experiences of sex(uality), with primarily young adults and their partners, but including parents and care practitioners (See Chapter 3). This chapter is divided into four sections. First, I examine the meaning of sex(uality) – what friendships, relationships, intimacy, and sex meant to the participants in their own words. Second, I explore what young adults told me about their relationship experiences and how this compared with the views of other participants. Third, I examine the barriers and the extent to which LLTCs impacted on, influenced, or limited young adults' relationship opportunities and experiences. I recognise that there are links between the three sections. In the fourth and final section, I discuss the significance of the findings. I begin with the participants' narratives about 'friendships', foregrounded by the young adults.

6.1 *The meanings of sex(uality)*

The specific definitions, concepts and components of sex(uality) and their relationship with holistic care (Taylor, 2012) were discussed in Chapter 1, Figure 1 and Appendix A. These components included friendships, relationships, intimacy, love and sex. During the interviews, I asked participants individually to explain what the separate components meant to them. I wondered if this might have any bearing on their opportunities and experiences to have an intimate relationship. My aim was to go beyond common understandings and to show the range of variations which emerged from the participants' views about what sex(uality) meant individually. I suggest that shared understandings about sex(uality) cannot be taken for granted and that the meanings of sex(uality), particularly for young adults with a LLTC, are unique to those individuals.

Friendships

During the interviews, young adults said that 'friendships' meant different things and included friendships with their peers, partners, families, carers, and health and social care practitioners:

"Most friendships are with my own age group. But because I've spent so much time in hospital and hospices, schools and colleges, naturally I am also friends with some care staff, teachers, not crushes and things like that, just friends."

(Mini, YAM)

"I regard my CNS [clinical nurse specialist] as a friend. She's treated me for a long time and I trust her as a friend but I respect her as my nurse too." (Lily, YAF)

Both Lily (YAF) and Vincent (YAM) spent a lot of time in their mothers' company, on account of their LLTCs:

"Mum is everything to me. She's my friend, my mentor, my guiding light as well as my mother. When she [mum] was very ill sometime back, I thought I had lost everything. I really didn't know how I would cope without her." (Lily YAF)

For Vincent:

"Although I have made some friends through university, my family are my friends, especially my mum. She has always been there for me, from birth, my diagnosis and throughout my life. She's a true friend." (Vincent, YAM)

For most young adults in this research, increasing isolation became an issue for 'friendships' once they left full-time education and spent more time at home, with limited peer contact, such as *Morris*:

"I don't have many friends near home but I have made some friends at the day centre and at Sunshine Hospice and I get on with my Dad. He's a friend. But I've lost touch with friends I made at school and college." (Morris, YAM)

Like *Morris*, other young adults welcomed the opportunity for peer group socialisation at the 'Transition into adult life weekends', provided by both *Sunshine* and *Terracotta Hospices*:

"The residential weekends at Sunshine Hospice are great. It's a good way to see friends, face-to-face; otherwise I would spend a lot of time alone at home." (Fiat, YAM)

"I love the Tuesday afternoon and weekend events that Terracotta organise – it's great to meet up and talk to friends." (Austin, YAM)

Some care practitioners described the role of the hospice friendship groups as providing opportunities for young adults to meet and socialise:

"We hold a monthly transition group here at the hospice specifically to enable young people to receive treatments, make friends and socialise face-to-face. Many young people have made friendships in this group and they meet up as friends during their short breaks here. Some try to maintain those friendships, mainly online, when they're not at the hospice. Meeting up with their friends away from here is tricky because of transport, distance etc." (Walnut, CPF, Sunshine)

“The great thing about the transition group is that they [the young adults] can spend time and meet up with their friends whilst having treatments at Terracotta.” (Peach, CPF)

However, for most young adults, friendships meant having a peer friendship, either with the same or opposite sex (for example, Lily, YAF, Jaguar, Alfa Romeo, Mini and Maserati, YAMs):

“Friendship... it is just someone that is interested in you, cares about you regardless of gender and whether you have any common ground or not.” (Lily, YAF)

Most of the young adults said that peer friendships did not usually include intimacy or sexual activity:

“I think friendships are with someone you can talk to about general things, how things are going. I’ve had a friend since age three. This was a male friend so I’ve known him for a long time. If you have known them for longer, then you build up trust, so I suppose that is what friendship is about, not sex and all that, that’s separate [pause] but someone you get on well with.” (Maserati, YAM)

Some young adults acknowledged that friendships could be with both the same and opposite sex:

“Friendships are important with both men and women. Friendships, they’re people you share your thoughts with, talk to, and have a laugh. Men and women are both equally important in friendships but it’s not really about sex.” (Jaguar, YAM)

Three young adults made a distinction between ‘friendships’ and ‘relationships’ (Alfa Romeo, YAM, and Lily, YAM):

“I have had lots of encounters and friendships with girls but no real relationship in my life, no girlfriend. These are just friendships with girls, not a relationship, as such.” (Alfa Romeo, YAM)

Although I specifically asked *Jane* to tell me what a friendship was, she compared friendships with relationships, sometimes to surprising effect:

“Friendship, right. I love my friends but in a different way to the way I love Dee [Jane’s boyfriend], but I think a friendship is more important than a relationship with your boyfriend.” (Jane, YAF)

In my second interview, I asked *Jane* why she felt friendships were more important:

“Well Dee’s just my boyfriend, not sure really why. It’s just different, can’t explain why.” (Jane, YAF)

In the absence of an answer, I questioned whether a friendship was needed and assumed to be part of her life:

“You need your friends. But with your friends you can go and talk to them about anything, you know. And that’s how it should be, that’s what good friends are for” (Jane, YAF).

Several young adults discussed the differences between face-to-face friendships (Morris and Jane, YAs), and those generated through social media (Austin, Maserati, YAMs, and Smidge, PF):

"I used to have friends at university and some still see me; but then there's the friendships I have made on Facebook but they're different. You don't see them in person, it's not the same, not as good. I enjoyed my university friendships because we met in person." (Austin, YAM)

Similarly, *Maserati* said that he would prefer to make friends with a girl face-to-face rather than via social media:

"Yes, I use the internet.... But I hate Twitter, Facebook, social media and all that stuff. I won't use internet dating or internet games. Friends on Facebook for me are unsociable. They are printed pages. Facebook requires constantly taking time, and it's not very sociable. I want face-to-face contact and friendships with girls, not pictures and the written word on the internet [pause]. I am quite conservative, traditional, so I would want to meet a girl face-to-face, to make friends before I have any relationship with her." (Maserati, YAM)

However, in the absence of meeting friends face-to-face, social media fulfilled a necessary role for several disabled people in both this research. Once young adults left either full- or part-time education, making face-to-face relationships became harder and their dependency on social media increased (*Lily* and *Marmaduke*, YAFs, *Austin*, *Vincent*, *Fiat*, *Jaguar* and *Lamborghini*, YAMs):

"I used to have face-to-face contact with friends but as my life-limiting condition has progressed, I see them less and have to rely on the internet. Without that, bar my mum and sister, I would be quite isolated." (Lily, YAF)

"Since leaving university, I spend a lot of time at home and rely on the internet to keep up with and make friendships." (Austin, YAM)

"I maintain university friendships via the internet and met Smidge [Vincent's fiancée] online." (Vincent, YAM)

Several parents, such as *Liliana*, *Goldie*, *Topaz*, *Crystal* and *Precious* (MFs) felt that social media filled a gap in the absence of regular face-to-face 'friendship groups':

"Friendships, Lily has made friends with people on the internet, both young girls and boys. She was friendly with, and sadly he died last year, a chap that she met through the Hospice and we'd bumped into him a couple of times, so she has got 'friendship groups', but a lot of them are not face-to-face friends and, we do try, if we go up to hospital and there's somebody in and we could go and visit them, if it's possible we will. So she does have friendship groups, but not necessarily at home, people that visit her, or events she attends provided by the hospice or the national charity [not named here]. But it's mainly online." (Liliana, MF)

While other parents recognised a place for social media, *Goldie* (MF) expressed her concerns about some of the internet friendship sites:

"Yes, my son, [whom I did not interview] does use the internet to make friends. He has a few friends at school. At this stage, I would have concerns if he used internet porn. I don't know if he does. I would be fearful if he used the wrong site and was deeply upset by what he saw there, such as violent or aggressive sex. But for making friends, when you're at home a lot, well the internet has a place without friends." (Goldie, PF)

Social media had enabled some friendships to be established and, in *Vincent's* case, to move on from 'online dating' to face-to-face intimate relationships. *Smidge*, his girlfriend, described that a benefit of meeting *Vincent* face-to-face was learning about his cystic fibrosis. This might not have occurred had they remained online friends:

"I met Vincent over the 'net' and we soon became friends. But I didn't know about his condition until we met face-to-face. I might never have known about his cystic fibrosis had we just remained internet friends." (Smidge, PF)

Overall, most young adults told me that they would choose face-to-face friendships in preference to those mediated via social media:

"Friendships... that's someone you can have a good time with. I have two close friends with cerebral palsy. I have known them for ten years. They are good mates, one male and one female. They attend the local respite centre with me so I see them a lot. That's important. So much better than meeting on the internet. I would choose face-to-face friendships any day." (Fiat, YAM)

Some young adults also said that the presence of their LLTC sometimes impacted on making friendships with their peer group, and were also influenced by gender:

"I don't think the girls were all that bothered by my condition and were happy to make friends. But I think the boys were more, yeah [pause], different. Definitely behaved in a different way in terms of friendships once they knew about my life-limiting condition." (Fiat, YAM)

Existing friendships sometimes changed once the individual knew about the young adult's LLTC:

"Yeah, people who I'd probably previously been friends with, didn't speak to me anymore and just avoided me [because of the LLTC]... There was definitely some of that. Where I probably would have expected to pick the friendship up again, it didn't necessarily happen." (Fiat, YAM)

Dee, Jane's boyfriend, who was non-disabled, described and highlighted choices which he was able to make about the how, where and when he could have a friendship, which for him was not exclusively at home:

"Friendship is being able to relax with somebody; at work, in the pub, the park, at home, whenever, wherever I want. It's more difficult for Jane because of her disabilities." (Dee, PM)

Austin's mother, Pear, explained that she could talk about her son's illness, as she did in Chapter 4, but found it difficult to talk about sex(uality) with her son:

"I think it's difficult to discuss the meanings of friendships, sexuality, relationships, love and intimacy when this mainly refers to Austin and not to me. I am very happy to fill in the gaps about his medical condition but find what he might perceive as friendships and all that stuff quite difficult to talk about with him." (Pear, MF)

Other parents were comfortable discussing their son or daughter's means of engaging with friends and making friendships (*Precious, Liliana, Topaz, Crystal and Daisy, MFs*):

"Having social contact with other people around her own age is probably at the top of her agenda, hence why she absolutely adores being at college. She'd rather be at college than at home, and has made it very clear she's not coming home as she has few friends here..." (Daisy, MF)

Daisy then went on to discuss how her daughter liked friends of either sex and from her own age group:

“She’s always been a very, very independent young lady. Even as a young child, she never wanted to be anywhere but busy with others of her own age, either females or males. They are all her friends.” (Daisy, MF)

Crystal spoke about the particular importance of friendships for her daughter who had communication difficulties:

“...because she can’t speak well. Marmaduke really needs to see and meet friends whenever possible and I really encourage this so that she’s not isolated.” (Crystal, MF)

Care practitioners described ‘friendships’ in relation to their own understanding and experiences rather than the young adults. Their views resonated with Dee’s comments about being able to choose when and how one meets up with friends:

“Friendship for me is somebody, like-minded people, people that connect; I can meet up with, someone on my wave length, loyalty.” (Ellie, CPF)

“My friendship experiences allow me to pick and choose when, how and where I meet up with someone. I am not constrained by physical limitations and disabilities and my friends, maybe my Mum, university pals, whoever.” (Peach, CPF)

Relationships

Most of the young adults said that relationships were deeper than friendships and could be with the same or opposite sex. Relationships were reported by some adults as an ‘amplification’ of friendships and included commitment and trust. At the time of interviews, five of the 13 young adults said that they had or were currently in a

relationship and these were (or had been) heterosexual relationships. No one stated that they were LGBT:

"It's trusting. For me it's, regardless of how you feel, always looking out and trying to take care of the other person. Relationships are like friendships but amplified and deeper." (Austin, YAM)

And:

"A relationship is a bit more than a friendship and yes, it's deeper. This relationship can be with either a boy or girl and may or may not include sex." (Jaguar, YAM)

For Smidge, Vincent's partner, it was the next stage, a commitment:

"Relationships require commitment from each other. For me, it's the next stage on from a friendship." (Smidge, PF)

Maserati talked about never having had 'a proper relationship', implying that he would like a girlfriend and companionship:

"Relationships... Hmmmm. These are more about the companionship side of friends. I have never had a proper relationship. I would like to have a proper relationship with someone, you get bored talking to yourself, and we all need someone. Yeah, a girlfriend would be nice. I would want to hold hands with her. Uuuhhhh. So relationships are not just about holding hands or having sex but can be...." (Maserati, YAM)

Several young adults told me that they were encouraged to have relationships with disabled people (*Mini, Fiat and Jaguar, YAMs*):

"We were encouraged to have relationships with disabled people. This did work out for me initially and then it didn't. I'd like to be able to choose." (Mini, YAM)

"I had a few relationships with girls in a wheelchair." (Fiat, YAM)

"Trying to see or visit someone in a wheelchair and make a relationship with that person is really hard in terms of travel, transport, cost and mobility." (Jaguar, YAM)

Lily described some of her difficulties in meeting non-disabled people, particularly in hospices, and drew attention to the heteronormative, 'ablest' cultural assumptions that disabled people want or need to be with disabled people (Liddiard, 2011, 2018):

"Relationships. I think people assume that a disabled person is going to want to be with another disabled person, but it is not always like that. There is no way that a young person, unless they go to college or university, will meet non-disabled people. Terracotta Hospice, that's fantastic, but I don't meet any healthy people there, they have all got a different condition, and as much as I want to be with a disabled person, I would also like to think that there is someone else who is non-disabled; so having the choice of whoever we want to be with, not you have to have this person because you are disabled which kind of, it is kind of geared towards you know, disabled people marry disabled people because healthy people wouldn't want to be with a disabled person. People find that quite hard, to think that a healthy person wouldn't want to be with a disabled person which is wrong, because you can love a person regardless of whether you are healthy or sick or poor or rich or whatever, it just depends on

the individual person but disabled people don't get a chance to meet healthy people and have relationships with them.” (Lily, YAF)

I interviewed *Diamond* (YAF) several years after her treatment for childhood leukaemia. *Diamond* described the loneliness of her teenage period when she was ill and said that she felt ‘isolated’. She talked about the importance of relationships to her now and what constituted ‘a new normal for her’ following her initial treatment for her LLTC condition:

“Relationships to me are everything. I need people around me and because I was alone as a teenager and I was ill, I wasn't socialising as I should have been, it had a big sort of impact on my own emotional wellbeing because I wasn't around people. I was isolated. So it's really important now that I feel normal again, to feel loved and to love as well, particularly after my illness.” (Diamond, YAF)

I asked parents what they thought relationships meant to their son and daughter. Some parents felt they did not know the answer, but others shared their hopes and expectations:

“Well, this would be a relationship with a girl. He [son not interviewed] hasn't as far as I know ever had a girlfriend but would love one and I don't think he's gay.” (Precious, MF)

“I would love Lily to have a friendship with a boy, someone who would love and respect her, for the wonderful person she is.” (Liliana, MF)

“I hope that my son [not interviewed] will have a relationship one day with a girl and that it's not just limited to the internet.” (Goldie, FM)

Intimacy

Intimacy has been described as:

‘The real taboo in our society – it’s the thing we fear, because it’s about taking off the mask that so many of us hide behind. But it’s the key to being freer, happier and more alive and it could change not only our personal lives, but the political decisions we take as a society.’ (The Guardian, Joanna Moorhead, 29 April 2017, accessed 7 December 2017)

Most young adults I interviewed said that intimacy required physical contact with another person, such as touching them, physical proximity, holding hands or a kiss (*Diamond, Lily and Jane, YAFs, and Smidge, PF*). It was at a ‘further level’ from a friendship and relationship and could include ‘having sex’:

“Intimacy... hmmmmm. It’s really important to me that we let each other know that we love each other, that we cuddle, that we have a kiss when we leave each other.” (Diamond, YAF)

“Intimacy, yes, it is, it’s a very touchy-feely, intimate relationship [with Vincent]. I think we are both like that as people anyway. And even when it makes other people uncomfortable. I think we are always touching, like if we are in the same room, we are generally always touching so I suppose when we are alone it is just a further level to that. We sleep touching so I chase him across the bed when he gets too hot. And we wake up on his side of the bed, I think nearly falling out. So I think intimacy it’s definitely sort of an attraction to always be close together.” (Smidge, PF)

For some young adults, such as *Jane*, intimacy was not solely about ‘having sex’ and included:

“...a nice cuddle. My love doesn’t have to be through sexual actions, not just having sex.” (Jane, YAF)

For *Austin*, intimacy included smell or touch:

“Intimacy is, for me, kind of that bond between a friend and a friend, or a girlfriend or a boyfriend, or same-sex relationship, if you go that way. It’s more – it doesn’t have to be physical, you know, you don’t have to have sex. It can be touching or smelling someone, but it can be having sex.” (Austin, YAM)

Some young adults, like *Maserati*, could not explain intimacy as it was something that he said he had not yet experienced:

“Intimacy? ... I am not really sure because it’s not something I haven’t really experienced [PAUSE] no I am not sure” (Maserati, YAM)

For *Smidge (PF)*, intimacy with *Vincent (YAM)*, her partner who had cystic fibrosis, also included phone and text communication, as well as being physically in contact with each other:

“We spend a lot of time apart because I work quite long hours, so we do speak a couple of times or at least once a day on the phone and never really hold back on our feelings, so we... Although he is quite awkward on the phone – you have to kind of liven Vincent up – but I suppose we are quite intimate in that way and we speak quite freely about everything intimately on the phone or via text when we’re not together during the day.” (Smidge, PF)

Similarly, *Crystal (MF)* spoke about *Marmaduke* and how she had previously used her communication aid and phone, to be ‘intimate’ with her former (and now deceased boyfriend), to express her intimacy:

“Marmaduke would send intimate texts to her now deceased boyfriend via her communication aid. She had a very strong attachment to one of the students at the college that Marmaduke attended [Zephyr was not a participant in this research]. He [Zephyr] was very caring about Marmaduke, and he would sit with her and hold her hand because she ate so slowly and wait for her to finish. And they would move their power chairs to the play area, you know, the playground or whatever and sit outside spending time together and being intimate in their own way.” (Crystal, MF)

At this point, *Marmaduke* cried as she recalled *Zephyr* and typed in some symbols into her communication aid, recalling their times together.

Sex

‘Having sex’ or just ‘sex’ is commonly used heteronormatively to mean ‘penetrative heterosexual intercourse’ (The Open University Sexuality Alliance, 2016; WHO, 2006a) and:

‘Disabled people are assumed to lack the bodily requirements to perform heteronormative sexuality and their alternative means of acquiring pleasure often remain unrecognisable.’ (Liddiard, 2011, p. 42)

Most young adult participants in this research described this term heteronormatively (*Diamond* and *Morris*, YAs, and *Jane* and *Dee*, *Lily*, *Vincent* and *Smidge*, *Lamborghini* and *Fiat*, YAs). In particular, *Fiat* commented that:

“Sex is something that is very complicated, it’s something that might improve the relationship, confirm what people think about each other.” (Fiat, YAM)

Austin, Jane and Maserati (YAs) acknowledged a distinction between ‘love-making’ and sexual intercourse, separating ‘the erotic’ and ‘the physical’ from ‘the romantic’. Maserati (YAM) expressed this separation as follows:

“There is a difference between sex and making love. Sex is having a good time for both parties, you know, both a man and a woman, [whereas] making love is romantic. They are not the same thing. Sex usually includes the act of intercourse, yeah [pause] that’s the difference; penis and vaginas and all of that anatomical and erotic stuff.” (Maserati, YAM)

With ‘sex’, relationships ascended to a different level for many participants and required ‘commitment’ (Smidge, PF), ‘a bond’ (Dee, PM), ‘trust’ (Diamond, YAF) and ‘unconditional love’ (Smidge, PF), or just ‘having sex’ (Jane, YAF).

Some participants, like Jane and Vincent (YAs), stated that they were ‘having sex’ or had previously ‘had sex’ (Vincent, Jane, Diamond and Morris, YAs), while others had not ‘had sex’ (Lily, Marmaduke, Austin, Fiat, Mini, Jaguar, Lamborghini and Maserati, YAs).

Cherry and Spruce (CPFs) indicated that sex might have other meanings:

“Having sex is very different for young people with a life-limiting or life-threatening condition; sex might just be something that you just watch on a computer screen as opposed to something that you physically participate in.” (Cherry, CPF)

Whilst Spruce (CPF) commented

“Well, some young people see sex only as internet porn, as not many have girl- or boy-friends and some aren’t that interested in having physical sex. Too much hard work with a ventilator or Percutaneous Endoscopic Gastrostomy (PEG)”, see Appendix A. (Spruce, CPF)

Other care practitioners saw a role for:

“Lap-dancing clubs or trained sex and therapeutic workers to alleviate [sexual] frustration are important if young people find it difficult to have sex.” (Walnut, CPF)

Lily explained how she felt her body could not ‘cope with anything really intimate’:

“I don’t think my body could cope with anything really intimate, but again just someone holding your hand, kissing you, hugging you, all sorts of things, it doesn’t have to just mean sex, intercourse and all that.” (Lily, YAF)

I suggest that the presence of the LLTC may have influenced the settings in which friendships and relationships were formed, while for others, the LLTC might herald the end of a friendship or a relationship. The different settings where young adults met may have influenced the nature of friendships, for example, facilitated friendships via social media, versus settings and circumstances where face-to-face friendships were maintained, such as ‘transition groups’, or where friendships ended, after leaving full-time education (Morris, YAM) or following the death of a boyfriend (Marmaduke, YAF).

6.2 *Relationship experiences: sex(uality)*

Overall young adults reported various relationship experiences. These included young adults who were in 'steady' relationships and 'having sex' with their partners (*Jane, Diamond, YAFs, and Vincent, YAM*), those who were or had previously been in a relationship, those who were not, (*Lily YAF and Maserati, YAM*), and those who used different forms of sexual fulfilment as alternatives to 'have sex' in the absence of a partner (*Jaguar, YAM and Focus Group, Sunshine Hospice*). These included: attending lap dancing clubs, internet porn, masturbation, or a combination of these. Three young adults did not wish to discuss sex and relationships with me (*Lamborghini, Jaguar and Mini, YAMs*). This section is divided into the following three sub-sections: those in relationships, those who were not, and those who used different forms of sexual fulfilment.

The restrictions and limitations imposed by a LLTC may have a significant impact, often making that transition to adulthood challenging (Abbott and Carpenter, 2012). Chronic ill-health may delay sexual or pubertal development and growth, and accelerate their neurodegenerative disorders. The young adult may have reduced opportunities to socialise with peers, and fewer opportunities to discuss sex(uality) with their peers and engage in relationships (Blackburn, 2002).

Experiences of sex(uality): those in relationships

Five of the 13 young adults said that they had been in, or were currently in, a relationship with the opposite sex, but the remaining eight said that they had not yet had a relationship with someone from either the same or opposite sex. *Lily* told me that she had not yet had a boyfriend. She discussed the impact of the LLTC on having a relationship:

"I haven't been well enough really to have a relationship with a boy. Again I haven't had time to meet anybody and have a relationship because of my constant ill-health. But everyone wants to be loved, don't they?" (Lily, YAF)

Of the 13 young adults with LLTCs, *Diamond* was married to an able-bodied man and had two children. Two other young adults (*Jane* and *Vincent*) were in long-term, sexual relationships with non-disabled adults of the opposite sex. *Vincent* was engaged to *Smidge*, his able-bodied girlfriend and *Jane* told me that she was not planning to marry:

"Marrying would be just too difficult. I don't want and probably can't have kids anyway." (Jane, YAF)

Care practitioners stated that most young people they care for wanted to socialise and have a girl or boyfriend. They talked about the role of social media as well as the importance of personal assistants in facilitating off or online relationships with young adults:

"Relationships? Well not a lot of young people want to say 'I desperately want a girlfriend or boyfriend and have a relationship.' We know that there is a social media that they [the young adult] from where can get a personal advocate or a personal assistant... we have a young man who lives alone with his mum and dad and has a PA who acts as a social assistant for going to the pub with him as part of his care package, and help facilitate with relationships, for example someone they can access to do things, that they cannot do themselves, accessing the internet, wanting to socialise." (Primrose, CPF)

Smidge and *Dee*, who were non-disabled partners in relationships with *Vincent* and *Jane* (YAs) respectively, both felt commitment and responsibility to their partners:

"I have been in a relationship with someone who has been very ill before. So I did not take on my relationship with Jane lightly. It's a big responsibility, not being able to see and her continence. I did think about all the complications of her condition but wanted this relationship with her. I had to commit, and we have sex." (Dee, PM)

"This is a relationship I don't feel like is something that I can easily or want to walk away from. I really want to be with Vincent." (Smidge, PF)

Morris had previously had a 'sexual relationship' with a non-disabled girl:

"I had a disastrous first sexual experience. I had a seizure during it and collapsed. It's put me off having sex ever since as I almost died." (Morris, YAM)

Of the remaining young adults, Alfa Romeo, Mini, Jaguar, Maserati and Lily had never had a boy or girlfriend but said they would like to have a heterosexual relationship:

"I would truly love to meet a guy, preferably face-to-face but even online would do." (Lily, YAF)

Both Marmaduke and Austin had previously had a boy/girlfriend but 'had not had sex':

"Yeah I have had a girlfriend, holding hands and all that." (Austin YAM)

Austin, at the time of his interviews, said he was in an online relationship with a young woman with a disability:

"I hook up with Hyacinth [not interviewed] online every day. We met online. She's a sort of girlfriend." (Austin, YAM)

Although no one volunteered that they were asexual, three men with DMD told me that they were ambivalent about their sex(uality) (*Mini, Jaguar and Lamborghini, YAMS*):

"I don't think I'm gay but I'm ambivalent as to whether I like girls or boys better. I don't feel particularly sexy." (Jaguar, YAM)

Maserati (YAM) suggested that although he was heterosexual, his cancer, at times, made him feel sexually ambivalent:

"The condition has impacted a lot on my life, my self-confidence. I am edgy, I have no libido... you know, sex drive? Libido – yes, the loss of libido – chemo and radiotherapy zaps energy levels... and some bodily functions... erections and all that and sometimes because of my condition, I feel I don't have a sex at the moment, I feel asexual, although in my heart, I know that I like girls, I don't feel sexual attraction to anyone at the moment." (Maserati, YPM)

Most participants were asked if they would 'like to have sex'. In addition to those who told me that they had already 'had sex' (*Diamond, Jane, Morris and Vincent, YAs*), *Marmaduke* through her Makaton communication aid said:

"Yes, I would like to, if I meet the right person." (Marmaduke, YAF)

Of those who had never 'had sex', two young people replied that 'they were unsure about "having sex"':

"I would love to have a girlfriend but couldn't cope with having sex at the moment, whilst having chemo and all the treatment... It would be just too exhausting." (Maserati, YAM)

Alfa Romeo (YAM), who has DMD, felt that managing his nocturnal ventilation or continence in a sexual relationship would require both advance preparation and assistance from others.

“As I use night-time ventilation... I would need to get the equipment into a correct position. I could probably talk about this with a woman but it might be off putting and a lot of work and preparation. Also not sure if my body could cope and I would need to ask my carer or hospice staff to help me and that opens up another can of worms which I’m not sure that they would be allowed to address.”
(*Alfa Romeo, YAM*)

At their invitation, I interviewed five parents of the young adult participants with LLTCs (*Topaz, Crystal, Liliana, Pear* and *Daisy, PMs*), and another five parents who were not related to the young adult participants. One parent (*Topaz, PM*) ‘thought’ that her son, Vincent had ‘had sex’ and ‘previous intimate relationships’. Seven parents stated that their son or daughter had had a non-sexual relationship with the opposite sex (*Silver, Goldie, Precious, Emerald, Liliana, Crystal* and *Pear, PMs*):

“I don’t think my daughter’s had sex, although she’s at college, I don’t see her all the time but I know she has had a relationship with a boy.” (*Daisy, MF*)

Three parents stated that their sons were in ‘online relationships and probably looked at porn’ (*Pear, Silver* and *Goldie, PFs*). Two parents thought that either their son or daughter would like to have sex (*Topaz* and *Liliana, PFs*) but expressed some reservations:

“Lily would not be well or strong enough to have sex because of her condition.”
(*Liliana, MF*)

"I know that Vincent and Smidge are very intimate and try to have sex but Vincent's body is quite weak." (Topaz, MF)

Goldie thought that her 16-year-old son (not a research participant) would be better:

"for the moment obtaining other forms of sexual relief such as masturbation."
(Goldie, MF)

Relationship experiences: those who had 'had sex'

Four of the 13 young adults had 'had sex with the opposite sex' (*Diamond, Vincent, Jane and Morris*). *Diamond* was married, and *Vincent* and *Jane* were in 'long-term' relationships with non-disabled people. In addition, *Morris* had previously 'had sex' with an able-bodied young woman. Of the remaining nine participants, five stated that they had never had any form of relationship, either heterosexual or same sex (*Alfa Romeo, Mini, Jaguar, Maserati and Lily, YAs*) but would like to do so. However, their lived experiences of 'having sex' varied. Not all the young people I interviewed said that their sexual experiences were positive. *Diamond* regarded sex as 'functional' but not really enjoyable:

"I don't really enjoy sex with my husband. It's all a bit functional." (Diamond, YAM)

Vincent and *Smidge* discussed the advanced preparation required before they could become sexually intimate with each other, for example having physiotherapy and treatments before having sex. Both *Vincent* and *Smidge* indicated how this was important for *Vincent's* well-being but inhibited spontaneity. Factoring in fatigue for *Vincent* after being intimate was also important. Similarly, for *Jane* and *Dee*, advanced preparation was required before 'having sex'. *Jane* was visually impaired and

sometimes required assistance in locating her anatomy (urethra) in order to self-catheterise before 'making love'. This was essential to avoid her 'weeing' on *Dee* during sex. Similarly *Jane* required someone to assist in removing her incontinence pads. Both partners, *Smidge* and *Dee*, appeared to be supportive regarding the necessary procedures required before 'having sex'. Taylor (2012), in her study about the sexuality of older couples with LLTCs, discussed the use of equipment and the importance of advanced preparation in respect of preparing for intimacy. Taylor also addressed the importance of rest and energy levels and their significance to minimise stultification of the couple's overall sexual experience.

Those who had not had sex

Three men with DMD (*Mini*, *Jaguar* and *Lamborghini*) described their feelings about sex(uality) as 'not sure about sex'. Now in their late twenties and early thirties, the same men stated that they had not had a heterosexual or same-sex relationship, felt that 'friendships and relationships were important' (*Jaguar*, *Lamborghini*, and *Mini*, YAMs) and that friendship was often a first step to something more:

"I made friends myself, somewhere to start from limits me as a non-able-bodied person. I hung out with girls. I was not necessarily confident with girls... I needed to be confident to make a friendship in order not be rejected... I wanted to meet people. I wanted to have a friendship and nothing more, didn't really want sex... I wanted to get out more... You know... getting a format for meeting people, making friends and all that." (Mini, YA)

Crystal (PM) described how her daughter *Marmaduke* was able to have a relationship with her boyfriend *Zephyr* (not a participant in this study) with the assistance of her Makaton communication aid:

“Zephyr [not interviewed] kissed you on your hand. He was very good on his talker, you know the communication aid, so they just clicked. They went to the young adults centre together. And Marmaduke went to the prom [end of term ball] with Zephyr. They texted a lot, the two: How are you? What are you doing? Things like that. Basically nothing. Nothing serious... he came to visit often. So they had quite a good relationship. His health deteriorated. Physically he became frail. And I think, to be honest with you, it came as a shock to all of us. We didn’t know he was that ill, actually. Then he died. It was a hard time for you [looking at Marmaduke]. Yes. It happened while the whole coming together of circumstances and as their relationship was blossoming [Marmaduke starts to cry].” (Crystal, PM)

None of the young adults volunteered that they were LGBTQ+, but all five men with DMD acknowledged that they were ‘shy’ and that they mixed more in male company. Two care practitioners alluded to some young people being asexual:

“I think some of the Duchenne’s lads struggle with their sexuality until they get older and feel asexual until their mid-twenties. Their sexual maturity comes later.” (Walnut and Cherry, CPFs)

And, as Alfa Romeo stated in a subsequent interview:

“There are not many women over age 18 years at Sunshine. Ummmmm. There are more severely disabled women and they don’t understand well. I would like to try and go out and hopefully meet women without a learning disability.” (Alfa Romeo, YAM)

Parents and care staff said that young people sometimes became infatuated with care staff. *Crystal (PF)* reported that:

“Marmaduke gets very excited when Chevrolet [not interviewed] is on duty. She finds him very handsome and flirts a bit.” (Crystal, PF)

Whilst *Mini (YAM)* volunteered that he didn’t ‘fancy’ his home carers who were mainly male, he felt differently about care staff at *Sunshine Hospice*:

“Some female staff at Sunshine are attractive and they turn me on but nothing happens. Even if we thought about it, just wouldn’t happen, breaking the rules. Some staff are younger than me, some staff are friends but nothing could or would happen but they have to be professional. The only opportunity I have to meet young able-bodied females is at the hospice.” (Mini, YAM)

“My carers are [aged] around 40. They talk to me, we get on well, but no I don’t fancy him or other boys for that matter.” (Alfa Romeo, YAM)

There were mixed views expressed by parents and care practitioners as to how they viewed sex and young people with LLTCs, as *Alfa Romeo* explained:

“Mum and dad... they would take me out if we planned it... and I wanted to go to the cinema with a girl... or to events... they would buy me gadgets if I wanted and they would allow me to go with girls if I wanted to have sex....” (Alfa Romeo, YAM)

6.3 Sexual satisfaction and relief

For those young adults who were not in ‘relationships’, social media, pornography and lap dancing provided an opportunity for a sexual experience or sexual relief. Young adults, parents and care practitioners reported that the young person ‘watched porn’ on their laptop (*Morris, YA, Magenta, CP, Terracotta Hospice, Primrose, FM*).

Several care practitioners in the two hospices also noted that there were ‘tight controls’ about where viewing porn could take place (*Magenta and Millie, CPs*). There were strict policies that were reviewed regularly in the hospices about what could be accessed. Care practitioners also discussed the importance of privacy, separate units for children and young people, to ensure that young children were not present when older adults were viewing ‘porn’ (*Magenta, Millie and Ellie, CPs*). However, care practitioners and parents expressed different opinions about the use of viewing pornography. Some accepted that ‘porn’ had a place for a person with a severe LLTC; others had concerns about the use of ‘porn’ on religious or cultural grounds:

“At least it gives him some relief and it’s usually done in his own room.” (Goldie, MF)

“The children’s hospice is separated from the young adult unit, and only the adult unit is permitted to access soft pornography in the privacy of young people’s own rooms.” (Spruce, CPF)

“Some of the staff aren’t easy about porn being available on laptops in the hospice, on religious grounds. I don’t mind or have a problem with it and we now have a policy about the use of porn in the workplace.” (CPF)

Several parents and care practitioners recognised that some young people, particularly those who had severe neuromuscular impairments, required sexual relief. Several staff and parents discussed ‘masturbation’:

“We can see that he gets sexually very frustrated but he can’t tell us that. He has erections. My husband has tried to help him relieve himself on occasions but I can see that this is a difficult issue for both the hospice and us to deal with as it may be seen as abuse.” (Precious, MF)

"My son can still use his hands so does masturbate regularly and sometimes makes himself sore. I sense he feels some sense of relief and satisfaction from this." (Goldie, PM)

Internet porn

Fiat and Mini talked about obtaining sexual relief via the internet and looking at soft porn but expressed embarrassment in doing so:

"Internet porn? Well... I used to go to my friend and would watch a film... American Pie... I heard noises and saw friends watching porn in his room. I started laughing. I see why people use porn...." (Fiat, YAM)

"I have used internet porn... it can be frustrating... I know I can't experience real sex.... I feel frustrated and can't do it and feel very embarrassed... I feel unsafe watching sex... I watch it to find out and see what happens... because I haven't experienced this type of stuff." (Mini, YAM)

"I tried the internet... told them I was disabled and then people went funny... I met someone on dateline and said I was disabled and didn't want to meet... hmmmmm... what would my family think about lap dancing...? Not sure, I think they wouldn't approve... they're very Christian." (Fiat, YAM)

Two care practitioners (Walnut and Millie, CPFs) both talked about the need for young people to be able to masturbate but there are dilemmas when they need someone to assist them with this:

"Until recently we haven't had a policy in place regarding masturbation. Staff get much exercised about the law and not touching a young person's penis or vagina.

Through the sexuality group, we have explored ways how the young person can be taught to self- masturbate in private when they are attending the hospice. But this presents some difficulties for some young people who cannot move their hands or rub themselves and staff worry about breaking the law.” (Walnut, CPF)

Sex workers and lap dancing

Young people and care practitioners also discussed lap-dancing clubs. The focus group of men with DMD discussed their visits to a local lap-dancing club. *Jaguar* described it as “expensive. I would rather spend my money on something else”. *Mini* said “it was a bit impersonal, a girl comes and sits on your lap for a few minutes”. *Lamborghini* said “it felt a bit cheap and was not the same as having a relationship” and *Alfa Romeo* summed up the experience, particularly noting that the lap dancers don’t have any feelings for individuals:

“Lap dancing... well you see people briefly... they are paid to entertain you.... For a short time... it costs me money... there’s no long-term companionship and they [the girls] don’t have any feelings for you.” (Alfa Romeo, YAM)

Mini chose not to go on the lap-dancing trips:

“I was invited but didn’t go on the lap-dancing trip... I think I would find it embarrassing... I wasn’t staying at the hospice at the time... I don’t know what it would be like in front of me.” (Mini, YAM)

However, the male focus group at *Sunshine Hospice* agreed that the lap-dancing club filled a void for some people:

“It’s better than nothing.” (Jaguar, YAM)

Whilst staff also recognised that the lap-dancing club filled a void, some staff expressed concerns about potential adverse media attention if a hospice was known to be promoting the use of lap-dancing clubs, particularly if hospice vehicles were used to transport young people to and from the venue:

“The Board knows that some of the young adults have attended the lap-dancing club and they have discussed the safeguarding and reputational issues related to this. We have a policy in place but some of the staff don’t approve of lap-dancing outings at all, especially if they are transported there in the hospice mini bus. Again, I don’t have a problem with the lap-dancing club.” (Walnut, CPF)

“Well, for me... personally, no sex surrogate, worker or counsellor would be useful... I am not sure what I want to read about sex....” (Fiat, YAM)

“Ideally I would wish to build a relationship with someone who grows to like me. I have Facebook pages of lightly clad women... no sex calendars... I understand how my mum feels and wouldn’t want to embarrass her.” (Mini, YAM)

6.4 Barriers to relationships

For young adults with LLTCs, their equipment and mechanical aids, such as wheelchairs and ventilators, created barriers to relationship opportunities and experiences. The impact of such barriers must be factored into the choice of having a relationship, as three young adults in the focus group discussion (*Fiat, Lamborghini* and *Mini, YPMs*) explained:

“The practicalities of two people who use wheelchairs just adds to the inconvenience and complications of having a relationship.” (Fiat, YAM)

“For a couple who are both wheelchair users, you need to plan, particularly if you require assistance for someone to lie you on the bed and have a cuddle. That can create difficulties and tensions for both young people and staff, particularly in residential settings.” (Mini, YAM)

And:

“I would like to have a relationship with an able-bodied girlfriend. It might be easier to see if you can manage physically together but only if my wheelchair doesn’t get in the way then, but my only opportunity for a relationship has been with a disabled girl.” (Mini, YAM).

Presence of a life-limiting and/or life-threatening condition

I argue that my data has highlighted that the meanings and relationship experiences differed among young adults and this hinged on the presence of a LLTC. These differences were influenced by whether the young adult had acquired a life-limiting condition, such as Duchenne muscular dystrophy (DMD), or a life-threatening condition, such as a cancer, during childhood or adolescence. I suggest that two factors were important: first, whether the LLTC was acquired or not, and second, whether the condition was life-limiting or life-threatening and if this impacted on the young people’s interpretations of meanings and opportunities for intimacy.

I interviewed five men with DMD in this research, who were then in their late twenties and early thirties. They told me that it was enough for them to have ‘*a good friendship*’, ‘*hold hands*’, ‘*have a kiss*’, and were not really bothered about ‘*sex and all that*’ (Fiat, Jaguar and Lamborghini). Only one man with DMD, said that ‘*he had had a relationship*’ with someone of the opposite sex and that this had ended because they lived too far apart geographically.

Walnut (CPF) at Sunshine Hospice described the distinction between the relationship opportunities and experiences of young adults with LLTCs under her care:

“Most of the young people would like to have a relationship, be intimate and may be have sex... the whole package... but most just want to be able to text someone... Or speak online. However... oncology conditions can be very different... in a relationship... letting go about different expectations. Here we have a lot of people with DMD... DMD is very physically debilitating and deteriorating... DMD lads are very different from people with other LLTCs... they are often less experienced, shy, and very physically incapacitated and having sex in between using a ventilator might just be too much of a challenge.” (Walnut, CPF)

Peach, a medical student at Terracotta Hospital, commented on the vast amount of complex equipment that young adults with LLTCs were required to use. She described how this might disrupt or even ‘turn off’ or prevent a relationship opportunity/experience:

*“So many young people are on ventilators and they’ve got catheters and their kind of rooms as they say, are almost like mini ITUs and type of thing as their need for care is so huge. How on earth do they find time to engage in relationships with all that equipment they have to use is **so** tricky.” (Peach, CPF)*

Several young adults discussed the impact that their LLTC had had on some of the physical aspects, causing disruptions and dislocations in their lives. Jane and Vincent both described the impact of their daily routines. Routines had to be planned and adapted. Jane described the difficulties of ‘managing her incontinence’ and toileting before ‘having sex’:

“If we need to stop to do the pump, we’ll stop to do the pump, you know. That’s amazing really because I didn’t have that, it was like right I’ve got to do that now. Don’t go to the toilet don’t. Go to the toilet now and make sure you don’t need to go. I don’t know when I need the toilet or not, do I? Particularly before having sex with Dee.” (Jane, YAF)

Jane described the difficulties with her diabetic insulin pump and its impact:

“When we first started [having sex] and I had to keep taking the pump off, stopping and taking my pump off, I was like, it got annoying, you know. And I was thinking ‘he’s not going to stay with me for long because this is no good’. But now it’s just, we just do it before sex, you know it’s part of me.” (Jane, YAF)

Jane succeeded in adapting, although the trouble inhibited their spontaneity:

“You just learn, don’t you? Like at the beginning, that didn’t work, so next time, or maybe a couple of times after, I’ll try it that way, you know. It’s like now sometimes, if I remember, I take my pump off before, you know, [having sex].” (Jane, YAF)

Sometimes the use of equipment and treatments also impacted on sexual activity and spontaneity. For Vincent, this included physiotherapy:

“I have to be pummeled and have physio before I can even consider having sex. (Vincent, YAM)

Vincent, Lily, Diamond, Jane, Morris and Maserati (YAs) all spoke about the ‘energy’ that was required when one is in an intimate, physical and sexual relationship:

“Even if I end up with a disabled person just because they understand more about my condition and the energy required to manage it... I just don’t know if I have the guts or the energy at the moment to focus on having a relationship.” (Lily, YAF)

“I had no libido, no sort of sexual desire, because I’d almost think I’d embarrassed myself or shocked myself as to what had happened. So that was it, so yes that was part of the reason why I just didn’t think about it, didn’t have any desire for, yes so that was, that’s like weird. Having sex, for me it’s really exhausting and requires a lot of planning.” (Vincent, YAM)

Although several young people spoke of their desires to live independently, the reality is that 11 out of 13 young people were still living with their parents, bar intermittent respite or short breaks provided by young adult hospices. All 11 were significantly dependant on their parents or carers for their physical care, as *Lily* described:

“My mum does most of my care like injections and my TPN, my IV medication because the CNS [clinical nurse specialist] only has limited time, she has not got much time with the other nurses, but they are not really prepared to do a lot of the bulk, they can do bloods every so often and they come and... but they don’t have the time to come in every day and do my TPN, so it all falls down to my mum.” (Lily, YAF)

The impact on partners

Two young adults were in long-term relationships; *Smidge* and *Dee*, *Vincent* and *Jane*’s respective partners. *Dee* was *Jane*’s 52-year-old partner. He had spent a lot of his life caring for others so although he needed to learn about the specifics of *Jane*’s rare LLTC, he appeared not to be phased by it:

"I looked after my mum and my ex-wife previously, they've all had serious illnesses." (Dee, PM)

But like *Smidge*, *Dee* 'sneakily' went home to look up the facts about *Jane*'s condition, without her knowledge. From their interviews, *Vincent* and *Jane* did not initially want their partners to know a lot about their conditions. I suggest that this was because they didn't want to risk losing their partners:

"No, when you first told me what the actual condition was I sort of sneakily went home and looked up on it, and was like oh, oh dear, oh you can do that, there you go. So I know but no it doesn't bother me at all." (Dee, PM)

Both *Dee* and *Smidge* recognised the importance of learning about their partners' conditions, particularly if they were planning a more meaningful and longer-term relationship:

"No, I think, I think as you get to know someone you learn a little bit more and learning about what she has and what it's doing it just another step in that process [pause]. Having a relationship with somebody with a heavy visual impairment we both know it could, I mean Jane could lose all the sight eventually anyway." (Dee, PM)

"We were having dinner out on our first date. I could see that he was hiding something. It was his pills. He initially tried to hide his condition from me but recognised that I had sensed something and he then told me he had cystic fibrosis. He hadn't mentioned this over the internet." (Smidge, PF)

Choices and protection from harm

Although people with LLTCs were constrained by the limitations of physical and cognitive impairments, these findings have highlighted that environmental, institutional and professional attitudes and responses, as well as the impact of the complexity of the physical conditions, often constrained choices, stifled independence and suggested a need for 'protection'. There is a balance between protection from harm and sexual autonomy. Access to information about sex(uality), support from sex counsellors/therapists and engaging in relationships and intimacy. This study has highlighted the importance of protecting vulnerable people from harm but not always 'bubble wrapping' them (De Than, 2015). Allowing young people to take some risks and thus enabling them to explore their sex(uality) in a safe and trusted environment may increase their confidence and sexual identity:

"The importance, the risks, the theory of any situations where that would be an issue and that as care workers directly with these patients we need to be very aware of it and where that line crosses and no I think it's really important. As I said earlier I think it's about safeguarding not just the physical, the emotional or the mental, we've got to take this as a holistic issue and, yes definitely, but I think writing a protocol for this kind of safeguarding is very difficult because you know it's hard even to put capacity in a pot, we try and we do our best in medicine don't we but that's hard enough but when you're looking at ages of 18 and above as an arbitrary number, and the maturity and the childlike behaviours in some specific conditions that go over 18 means that 18 is not a relevant number for that whereas in the law it's x, y and z and for capacity you can say x, y and z but actually we have to be more individual case specific I think in these areas."
(Ellie, GP)

6.5 Discussion

During interviews the young adults, partners and parents simultaneously shared their hopes, aspirations and experiences for the young adults' relationships. Although I tried to separate the questions about the meanings from those about relationship opportunities and experiences, many combined their answers (*Lily, YAF, Vincent, YAM, Liliana, MF and Goldie, MF*). I argue that there is an interface between meanings and experiences. Indeed, the meanings and experiences were often entwined and inextricably linked together. I have studied the data to explore the ways in which 'the meanings and experiences' were embedded in participants' own words within their biographical stories (Plummer, 1995). The meanings and experiences of sex(uality) were often predicated upon and coloured by the presence, symptoms, treatments and concerns about their LLTC and the limited opportunities for young adults' social interactions with their peers. This discussion is addressed under three key themes that arose from the findings: the meanings of sex(uality), the young adults' relationship opportunities and experiences, and the barriers to relationships and sex.

The meanings of sex(uality)

I separated the sex(uality) interview components into five parts: friendships, relationships, intimacy, love and sex. Participants did not wish or feel confident in discussing 'love' as most of the young adults said that this was not something they had yet experienced (*Lily, Maserati and Fiat, YAS*). As outlined in Chapter 2, recent social scientific research (Abbott et al., 2016; Abbott, Jepson and Hastie, 2015; Abbott and Carpenter, 2014; 2012) have indicated that there are significant challenges associated with planning for and living life as an 'unanticipated adult' with a LLTC. These include being able to talk about friendships, relationships, love and sex, in the context of living with both the physical and emotional aspects of a LLTC. As young adults are now increasing in numbers as 'unexpected survivors', such matters have not always been

fully explored, for fear of upsetting family members, as well as the potential taboos often associated with both sex(uality) and end-of-life discussions. (Abbott et al, 2017).

Pubertal changes in early 'normal' adolescence may be characterised by rapid growth, mood swings, biological drive, socialisation and peer group identification. During mid-adolescence, peer group identification and risk-taking usually increases, authority and parents are frequently rejected, and sexual development and sexual activity begin. By late adolescence, young adults are more autonomous, independent and considering their life-styles, careers, relationships and finances, and are often moving away from home (Thomas, Bax and Smyth, 1989).

People with LLTCs have a different 'lived life experience' which maybe isolating, because they are not experiences that non-disabled people commonly share. I argue that *Smidge, Vincent's* fiancée, was an exception as no one had a physical disability in her family. *Dee's* first wife had had a life-threatening condition and Dee had also helped support and care for other family members. Having a LLTC may be isolating because of the pain and treatments but also because of feeling disempowered beyond the individual's immediate circle of support, and some non-disabled people not wanting to share or avoiding socialisation.

It is evident from this research that young adults with LLTCs often socialise with others who have LLTCs. Partly this is the comradeship of shared experience, for example, men with DMD may associate with men who have DMD. They do not have to explain what it feels like to have their condition. They are accepted and sharing will enable and facilitate friendships.

All groups of participants, particularly the young adults, acknowledged that the components of sex(uality) meant different things to each of them. The conceptual meanings and experiences of relationships, intimacy, love and sex varied enormously in this research (Liddiard, 2016; Maas, Shearer and Gillen, 2015; Liddiard and Goodley, 2015).

Relationship opportunities

In this research, 'being accepted' was the start of friendship. For many people, a friendship is the first step on the way to a relationship (Blackburn, 2002). What separated young people's 'friendships' for participants in this research were the lack of opportunities 'to connect' easily, particularly face to face with friends.

As noted in the previous section, friendships are usually built upon shared experiences – but this maybe more problematic for a disabled person. Having a LLTC is not an everyday experience. It is unique to the individual and many non-disabled people do not want to be part of or share those experiences. Having an LLTC is isolating. The presence of a LLTC demands time and energy that non-disabled people may except as good will but find hard to understand (*Lily, YAF*). It was more common for people with LLTCs to make friends with each other, for example, (*Marmaduke, YAF, Fiat and Mini, YAMs, Daisy, FM*). The LLTC also influenced the setting in which friendships were formed. The onset of a LLTC generally affected the nature of on-going friendships, sometimes heralding the end of a friendship (*Maserati and Diamond, YAs*).

Experiences of sex(uality)

Having a relationship meant different things to the young adult participants. Some participants wanted to have close friendships and relationships (*Jane, Vincent, Morris, YAS*), other young people wanted a girl/boyfriend (*Lily and Marmaduke, YAs*); some

had had a girl or boyfriend (*Marmaduke and Morris, YAs*). Some young adults wanted to experience intimacy and sexual relationships (*Jane, Vincent, Diamond and Morris, YAs*), whereas others were uncertain (*Lamborghini and Jaguar, YAMs*); some were happy to engage in online sexual and non-sexual relationships (*Austin and Maserati, YAs*), and others were thinking about the feasibility of having children (*Vincent and Smidge, YA and Partner*). Friendships for non-disabled young people usually develop before puberty and transition and are not necessarily hampered in the same way by the environment in which they live, work or socialise (Knox et al, 2011).

Participants acknowledged that sex and relationships were important to them. They also spoke about parents' and carers' roles in facilitating this. The young people frequently depended on others to facilitate sexual agency. Participants spoke about being recognised as sexual beings and as people with sexual requirement.

People with LLTCs often struggle to be seen as sexual beings with sexual needs, although participants in this research were clear to articulate the significance of sex and relationships in their lives. Sexuality is not seen as an integral part of the lives of disabled people (Liddiard, 2018, 2016; Taylor, 2012). Young adults discussed the way that an uncertain and shortened life course limited their opportunities for sex and relationships because other people assumed that this issue was neither relevant nor appropriate.

The young adults with LLTCs in this research were socially isolated and had limited opportunities to socialise. They had completed their education and training, had limited opportunities for employment and further training or voluntary work, and consequently spent a lot of time at home not engaging with their peers and 'without meaningful activity' (Abbott and Carpenter, 2014, p.192).

I suggest that men with DMD were less experienced in intimate relationships than people with other conditions such as those I interviewed with cancer and cystic fibrosis (*Diamond, YAF and Vincent, YAM*). They acknowledged that they were very shy (Abbott and Carpenter, 2014).

The findings highlighted that people with LLTCs often struggled to be seen as sexual beings with sexual needs, although participants were clear to articulate the significance of sex and relationships in their lives. As Earle (2007) has previously argued, sexuality is not seen as an integral part of the lives of disabled people. Young people discussed the way that an uncertain and shortened life course limited their opportunities for sex and relationships because other people assume that relationships and sex are irrelevant.

The barriers

The findings in this research have highlighted a number of barriers regarding sex(uality). These included the risk of physical harm from treatment and abuse, physical barriers, and the emotional experience of living with a LLTC and the fear of its return. A distinction was noteworthy between participants who had survived a LLTC and people whose health continued to deteriorate across the illness trajectory (Glaser and Strauss, 1965). The long-term implications for *Diamond* surviving Acute Lymphoblastic Lymphoma (ALL), a childhood leukaemia, reminded her of the ‘invasive treatments’ she underwent during childhood. *Diamond’s* “constant fear of recurrence” of “any cancer” (*Diamond, YAF*), the potential impact on her bodily changes and her own perceptions of femininity and sexuality as an adult resonated in the study of women following breast cancer treatment (Trusson, Pilnick and Roy, 2016), discussed in Chapter 2.

The young adults in this research surpassed clinical expectations. They had not only survived into young adulthood, but beyond, into their late twenties and thirties. Reaching adulthood was not an anticipated milestone for these young adults or families and therefore their transition to adulthood and, in particular, their opportunities to experience relationships, and the obstacles that they encountered on that journey', were both challenging and were not acknowledged (*Lily, YAF and Vincent, YAM*); as Giele and Elder (1998, p.22) have noted 'a sequence of socially defined events and roles that the individual enacts over time'. The findings indicated that people with LLTCs were sometimes infantilised by others and that sex and relationships remained unseen as an important marker of the transition to adulthood for these young adults (Liddiard, 2018).

Many of the young adults felt that they were living on borrowed time and felt damaged because of their LLTC (*Diamond and Jane, YAFs and Vincent*). Similarly, Kelly et al (2015) captured people's understanding of the impact of cancer on individuals, recognising features of embarrassment, for example, men feeling different from other men, like damaged goods.

The young adults in this study were experiencing progressive and deteriorating conditions, impacting on their health and social wellbeing during their adolescent development (*Jaguar, Fiat, Lamborghini, Marmaduke and Jane*). For other young adults, the experience was slower, and with their adolescence began the life-long dependence on others (*Vincent and Marmaduke*), notably families or carers, either at home or in residential settings (*Goldie, Precious, Topaz and Liliana, MFs*). The lived experience of 'borrowed time' created uncertainty and disruption, and forced the following barriers for young adults and their supporters: living with the potential side effects from treatment, the emotional changes of living with the LLTC, fear of its return or its exacerbation, and the differences between participants who have

survived a life-threatening illness and whose health was continually deteriorating, on an uncertain illness trajectory.

Transition was seen and remains a complex and challenging time, in part because many families had not expected their offspring to live long enough to grow into adulthood. As well as socially constructed barriers, young adults faced some limitations due to the severity of their LLTC, but also because of problematic assumptions about the nature and value of lives that were likely to be shorter than many others (Abbott et al, 2016) and the meanings and experiences of sex(uality), which were often devalued in these young adults.

Finally, these findings have highlighted that the ability of an individual to make choices throughout an uncertain life course may be constrained by a range of issues. The participants struggled with being seen as adults, and thus as sexual beings with sexual rights and needs. Sex(uality) and relationships were viewed as an important part of adulthood, although this was not always perceived as such by significant others in their lives.

6.6 Conclusion

The meanings and the experiences of relationships, intimacy were important to young adults with a LLTC but their opportunities and relationship experiences differed. These were constrained by several factors. These included whether the young adult had acquired a condition at birth or during early childhood, or in late childhood or adolescence, their opportunities to meet with their peers with LLTCs, once they had left school or college and limited opportunities to socialise with their non-disabled peers. One participant had had children and one man (*Vincent, YAM*) was considering having children. Genetics, fertility and reproductive opportunities are the focus of the next chapter.

Chapter 7 Uncertainty, reproductive hopes, choices and loss

Introduction

In this chapter, I focus my findings on the uncertainty about reproductive loss, the aspirations and hopes for reproductive opportunities in the future for the young adult participants, and the views of their partners (*Smidge, PF*, and *Dee, PM*), parents and care practitioners. First, I consider the uncertainties that participants discussed about puberty and adolescence associated with their LLTC. Second, whether the young adult, and the partner and parent participants had received or were offered genetic or fertility advice. Third, I address the participants' reproductive choices, opportunities and future aspirations, including their wishes to have children. Finally, this chapter draws together the themes from the participants' evidence to arrive at tentative conclusions.

Reproductive loss was discussed in section 2.3. Earle and colleagues (2007) have organised reproductive loss into three categories. I have adapted their organisation for my own findings in sections 7.1 to 7.3 by adding a fourth category, see Table 6. My focus in this research was mainly on the fourth category but there are examples of relevance from the findings in the first two. While the findings in this research cut across several types of reproductive loss, those with LLTCs and their consanguineous (blood) relatives form one group of people with particular reproductive considerations on a much larger spectrum of reproductive experiences. For people who cannot conceive, their lives may become increasingly medicalised and disrupted, and those with LLTCs are no exception.

Types of reproductive loss (medical or socially constructed).	How this type of loss presents	Participants identifying with this type of loss
Category 1	Where individuals may wish to parent a child but may be unable to do so without medical intervention. Experiencing and encompassing the loss of (or undermined) sense of reproductive identity, as well as the loss of an imagined child or family. (Graham et al, 2012: 207)	
Infertility	Loss of reproductive identity.	<i>Possibly several young adults</i>
Unsuccessful assisted conception	Loss of an imagined child.	<i>Possibly several young adults</i>
Repeated early miscarriage	Not volunteered in this research.	
Category 2	Encompasses a wanted pregnancy when the loss / event took place. These types of reproductive loss may involve the loss of an established parent status, alongside the loss of an anticipated future healthy child. (Graham et al, 2012: 207)	
Early/ late miscarriage	Loss of imagined, healthy child.	<i>Topaz (MF)</i>
Stillbirth	And loss of parent status.	<i>Not volunteered</i>
Neonatal or childhood death	Loss of parental and loss of a child.	<i>Goldie (MF)</i>
Termination for medical reasons		<i>Topaz (MF)</i>
Category 3	Not relevant to research participants.	
Termination of pregnancy for non-medical reasons	Loss of pregnancy and possibly loss of imagined child.	<i>No participants in my research</i>
Category 4	The loss occurs before planning reproduction, possibly on medical advice. It is a possible emotional loss of identity, although different from category 1, where reproduction may be attempted, even in the case of infertility.	<i>Lily (YAF), Vincent (YAM) and Smidge (PF)</i>
Early (precocious) puberty or later onset of puberty	Possible loss of reproductive identity.	<i>Marmaduke, Austin and Maserati (YAMs)</i>
Genetic implications	Possible loss of reproductive identity. Possible loss of imagined child.	<i>Vincent (YAM) and Smidge (PF), Diamond (YAF and PF)</i>
Treatment implications	Possible loss of parental status.	<i>Lily, Vincent, Maserati, Austin, Maserati (YAs)</i>
Uncertainty about Degenerative LLTCs	Possible loss of reproductive identity. Possible loss of imagined child.	<i>Lily, Vincent, Diamond, Austin (YAs), Pear and Daisy (MFs)</i>

Table 6: A typology of reproductive loss organised in four categories, three of which are adapted from Earle et al, 2007, p. 28 and Graham et al, 2012, pp. 206–207, while the fourth is a new category emerging from these research findings.

7.1 *Uncertainties about puberty and contraception*

Adolescence was a starting point in my research design (see Chapter 2 section 3). Puberty forms a natural starting point to conceptualise the findings that have contributed to understanding reproductive loss across the life course of participants in this research. Puberty, the biological marker of adulthood, heralding reproductive capacity. In certain societies, puberty triggers specific ‘rites of passage’ that formally recognise the transition from being a young person to a young adult.

Puberty, in the context of young people with LLTCs often required careful management. For example, immunosuppressant therapies may impede sexual functioning, such as erections (*Maserati, YAM*) and cause pain and discomfort in people with LLTCs and period discomfort (*Lily, Jane YAFs*). Several parents told me that their children were not expected to reach their teens (*Goldie, Precious* and *Crystal, PFs*) so subjects such as menstruation, erections and reproduction were not on their ‘radar’ (*Pear, FM*), as they had expected their son or daughter to die before adulthood.

For some young men with LLTCs, puberty may or may not signal the onset of early morning erections and wet dreams, resulting in the release of semen and emissions from the penis. When Goldie’s son masturbated and ejaculated, the “big mess” was cleared up by his mother (as the young adult could not ‘clean up after himself’, (*Goldie, MF*).

For young women, periods – and particularly the first period, the menarche – are usually significant milestones, signalling reproductive capacity. Menstruation was often perceived by participants as ‘messy’ (*Walnut, CPF*) ‘painful’ (*Lily, YAF*) and ‘embarrassing’ (*Jane, YAF*), and that young women needed help from others in changing sanitary or incontinence pads.

“You know? I wear incontinence pads and sanitary towels which he [Dee] knows about and helps me change, which is fine.” (Jane, YAF).

Jane required help with self-catheterisation because she was visually impaired (Jane, YAF).

“So to self-catheterise myself, and this, there’s, I have to get it in the right hole down there and I didn’t realise but until I started having sex, I can’t tell which one because it’s got bigger so I don’t, you know, so no-one got me ready for that and I’ve had to do it myself and I can’t see well. You know so there’s no bladder person that said look as you get older in puberty, this is what’s going to happen” (Jane, YAF).

Other participants described their experiences of puberty and adolescence, such as periods and menstruation (Lily and Diamond YAS, Crystal and Daisy, MFs, and Cherry, CPF).

“My bladder has failed, my blood vessels don’t constrict properly and it causes a number of different problems, including my terrible periods and when and how these occur. This is deeply unsettling.” (Lily, YAF)

In Lily’s case, there was uncertainty around her diagnosis, (discussed in Chapter 4). The physical effects of her LLTC and its treatment interrupted, delayed, and impacted on her bodily functions and pubertal development.

“I’m not sure I have the energy and the time to devote to someone else, with my care being so time-consuming. Do I have the energy after everything I have to do to have a relationship? I think if I could get my health more, kind of, under control... I could get out there with more people of my own age. But the

moment I'm mostly bedbound, and I'm not enjoying my late adolescence.” (Lily, YAF)

Since her cancer, *Diamond* had experienced pain and discomfort with her menstrual periods, and had questioned her ability ‘to conceive’ without fertility treatment:

“Yes. I had quite a lot of problems with my periods in my teens and twenties and when I was in my 20s, I wasn't having very regular periods, they were very painful and I was bleeding a lot. So I had a laparoscopy. When I came round from the laparoscopy I was seen by a couple of SHOs [Senior House Officers] before being sent home, who told me that I had polycystic ovaries and that I was unlikely to be able to conceive without fertility treatment.” (Diamond, YAF)

Diamond's previous life-threatening illness (cancer) added layers of complexity to a life which I argue had been held in flux/ abeyance about *her* fertility after her childhood cancer treatment. *Jane* (YAF) told me that she was living with and ‘having sex’ with her male partner, *Dee*. *Jane* had been prescribed injections of Depo-Provera by her GP to reduce the risk of pregnancy:

“Because I'm on the injection, you know, Depo-Provera, for my periods, I am not getting pregnant but now I don't have regular periods anymore. But I am still having sex regularly.” (Jane, YAF)

When I asked *Jane* whether she had been consulted about the use of Depo-Provera, she replied:

“Definitely, and I thought this best as I can't always see [Jane was visually impaired] or remember to take my [contraceptive] pills. My GP prescribed my

treatment. Hospice staff talked to me about it first. They were worried I might get pregnant without it.” (Jane, YAF)

During one of my three interviews with *Jane*, I asked her to tell me more about her periods. As this question was met with hesitation, I did not press her on this point. *Walnut*, her clinical nurse specialist, commented:

“I don’t think and I hope that Jane’s not pregnant. She’s not ready or well enough to care for a child yet and they [Jane and Dee] have not known each other that long either so we explained the benefits of having the ‘Depo’ [Depo-Provera] injection or she was OK with this.” (Walnut, CPF)

Walnut also raised some potential concerns about *Jane*:

“I recognise Jane’s visual difficulties ‘to take or find my tablets’ [Jane’s words] and her own rights to choose whether or not to have these injections [Depo provera]. But we had a number of concerns about her age, Dee’s age, safeguarding and all that. Though Depo was much easier for her, it might disrupt her periods but also stop her getting pregnant and make her periods a bit less messy which she does not manage well by herself.” (Walnut, CPF)

Walnut had tried to assist *Jane* in making an informed choice about the benefits of Depo-Provera. While involuntary sterilisation of disabled women, and particularly those with intellectual disabilities, is no longer practised within Western societies, the prolonged use of long-term contraception, such as Depo-Provera, may be viewed in some circumstances as a form of social control and a modern method of chemical sterilisation (Tilley, Walmsley and Earle, 2012).

Other participants commented on the dearth of information about managing periods (*Crystal, PF, and Marmaduke, YAF*). I asked both *Marmaduke YAF* and *Crystal PF* about the information they had received about puberty and sexual development. *Crystal PF* responded to my question in the presence of both *Marmaduke* and *Marmaduke's* nurse, *Aqua Marine CPF*.

"Maddie was asking whether we had any guidance or support when we [i.e. Marmaduke] reached puberty, development, about periods and sexuality and things like that and where I would have gone for help. I had nothing whatsoever as a mum for Marmaduke. Well, I don't really know. Apart from asking you, Nurse Aqua Marine, about periods and such like." (Crystal, MF)

I suggest that this raises issues regarding parental access to information about menstruation and periods and this aspect was detailed in Chapter 5.

Daisy described her daughter's periods. (I did not interview her daughter):

"My daughter was about 12 [when she started her periods] but then everything stopped again because she was so unwell. And then it started up again [her periods] but probably not until she was about 14. Oh, she was quite put out by her periods because she self-harms a little bit so normally she causes blood to herself. But my view is if everything's fine, [the periods are] not causing any distress or medical problems that don't interfere, that's my view, and you just have to manage what you have to manage." (Daisy, PF)

When I probed further about the support her daughter had received in relation to her periods and self-harming, *Daisy* explained:

"I know that she had some lessons with the school nurse to do with puberty and periods but I've no idea what was said, how it was said and when it happened. Or how she would manage her periods. We weren't, as parents, we weren't included [by the school in these discussions]. I have been included in discussions regarding C's self-harming and that's helpful. We've struggled to really pinpoint a pattern with her because she's very mood-swingey anyway and she has high anxiety and she can just switch literally from being sweet and lovely to being, you know, really quite cross with life very quickly. Certainly, I haven't noticed that it's more so at that time... you know periods etc." (Daisy, MF)

At a time when many adolescents seek independence and become more autonomous, people with LLTCs, like *Daisy's* daughter, faced a greater dependency on others. I suggest that *Daisy* was trying to resist this on behalf of her daughter, Whilst *Daisy's* daughter may have required assistance to manage her self-harming, and managing her menstruation may have been perceived as an invasion of her daughter's physical privacy (Brown and Sourkes, 2006). When I sought her views about the use of Depo-Provera, *Daisy* replied:

"I don't think [Depo-Provera] should be used for convenience for the parent or carers. I think it should only be used if there are problems, which is the balance. Because although these sort[s] of medications, they don't come without their own side-effects, so it's about balancing which is the best, really. But I wouldn't want to see somebody left suffering if they needed medication but my daughter would need to be assisted in making the right decision." (Daisy, PF)

I asked the young adult and parent participants about sex drive (libido), erections and masturbation. Some female participants did not wish to discuss this topic (*Marmaduke, Diamond YAFs and Crystal, PF*) whilst others were happy to (*Precious PF*).

“I think my son had PHSE, biology, general education... and stuff at school but we would really have appreciated more about how to manage masturbation, wet dreams, erections and none of that was covered and is important for us as parents too, especially now.” (Precious, PF)

Some of the participants told me that their libido was inhibited by their medications and treatments. (*Maserati, Morris and Vincent, YAs, Emerald and Goldie, Parents, and Oak and Ellie, CPs*). Although *Maserati* appeared shy and embarrassed during the interview, he explained that:

“The condition [cancer] has impacted a lot on my life, my self-confidence. I am edgy, I have no libido, and you know, sex drive. Libido – yes, the loss of libido, chemo and radiotherapy zaps energy levels and some bodily functions, erections and all that in my case.” (Maserati, YAM)

For *Maserati*, his cancer had impacted on his sex drive, self-confidence, some of his bodily functions and self-image. Libido may be decreased by drugs that block dopamine or testosterone, or that cause dysphoria. Erections may be decreased by drugs that divert blood flow from the penis, or drugs that affect spinal reflexes (NHS Inform, 2019), some of which may be used by people with LLTCs.

Morris explained that he had erections and recalled his negative sexual experience (when he had a seizure whilst having sex, discussed in Chapter 6).

“I still have erections when I see a pretty girl but I am frightened to ask a girl out. I have never really seen or rather been out with a girl properly since that devastating accident.” (Morris, YAM)

Although I asked participants about erections, wet dreams and ejaculation, there was a certain reluctance to discuss these matters, with the exception of *Maserati* and *Morris* (YAMs). I raised these issues because in my previous research (Blackburn, 2002), I asked male participants about the presence and absence of erections and young men volunteered this information. I was therefore surprised at the reluctance to discuss such topics in this research but accept that there may have been several reasons for this. First, in my previous research, I used a mixed methods approach: both face-to-face interviews and a survey questionnaire; thus writing down responses may have been easier and less embarrassing than talking about erections directly in interviews. Second, my gender and being an older woman conducting face-to-face interviews on a sensitive subject may have been a deterrent (Browne, 2005). Finally, I am older than when I carried out my previous sexuality research (Blackburn, 2002) and this may or may not have been a factor. I came to academia as a very mature student, following a professional career working in policy and practice addressing the needs of children and adults with disabilities (see Chapter 1.3)

Sometimes parents were more forthcoming about these topics than the young adults (for example, *Goldie* and *Emerald*, MFs). Parents mainly spoke about managing the practicalities of masturbation and ejaculation. *Goldie* described her son's experiences:

"Well because he makes a mess in his bed and that creates more work for me, Zephyr [not interviewed] has to ask me to please clean the bedding or give it an extra wash. He started masturbating when he was 14 and because I can talk to him, we don't make an issue of it. Not sure how I shall manage these matters in the future. We shall go on Duchenne family holidays if and when he is over 18 years and we shall at least be able to share these experiences with others. It is a normal human thing." (Goldie, MF)

Goldie was looking ‘to share these experiences with others’, recognising the normality of her own and her son’s experience, but she was also highlighting the isolation that some parents encounter in talking about ‘taboo’ subjects within ableist cultures (Liddiard, 2015). Liddiard (2011) noted that the male personal assistants were less likely to offer assistance with masturbation to disabled women, than female parents and personnel were to disabled men. Liddiard argued that this was:

‘Based on constructions of disabled women as vulnerable to sexual abuse and constructions of male personal assistants as abusers.’ (Liddiard, 2011, p. 287)

Emerald spoke of the dilemmas which both she and her husband encountered in managing their son’s sexual frustration *and* described ways that she and her husband used to alleviate *Cortina*’s frustration (neither *Emerald*’s husband nor her son were interviewed):

“It is about managing our son’s sexual frustration. If I put his [her son’s] own hand on his genitals, he calms down, but my husband can’t do this. He is concerned that he [my husband] might be thought to be abusing my son. My husband and I have discussed this a lot. Anyway, Cortina [her son] has facial hair. He has early morning erections when he appears frustrated, he rubs himself but I don’t think this is really linked to any sexual feelings.” (Emerald, MF)

Although, disabled people may be more susceptible to sexual abuse than non-disabled people (Gillespie-Sells, Hill and Robbins, 1998), their carers and supporters are also susceptible to accusations of sexual abuse and may find it difficult to engage in intimate care procedures, leading to perceptions of inadvertent neglect (Earle, 2001).

The effects of illness and its treatment on physical appearance also influenced how young adults' developed a sense of self, as *Maserati* described:

"I have a shower, make myself clean and presentable. As I don't always feel good about myself, no hair, eyebrows, pale skin but I'm different. I am not typical even with cancer, I'm not your typical lad from Brushire [fictitious name] where young men are conscious of their appearance and I don't want them to see me looking like this." (Maserati, YAM)

For most adolescents, physical changes during puberty bring about heightened concerns with body image (Blackburn, 2002). Thus, visible markers of illness, including, for example, hair loss, as in *Maserati's* case, places the adolescent at increased risk of poor body image and feelings of inferiority and low self-esteem. Compounded with *Maserati's* own difficulty in adjusting to an altered body image was the fear of others' reactions to his physical appearance (Browne and Sourkes, 2006).

I asked two general practitioners (*Ellie* and *Oak*, CPFs) their views about precocious (early onset) puberty in relation to people with LLTCs. Both *Ellie* and *Oak* were also hospice leads. They talked about their and others' responsibilities in addressing this:

"My impression of precocious puberty [see Appendix A] would be that those issues would be probably addressed by the consultant pediatrician in charge or their GP, rather than a hospice lead such as me. Therefore when these children would come to the hospice for respite care etc. that would be then something that the parent would hand over to the nursing staff and it [precocious puberty] would be handled appropriately and I, well, I've never been involved in it in terms of the hospice." (Ellie, CPF, GP)

Sometimes a LLTC may precipitate either early or late onset of puberty, such as the onset of early periods, facial and underarm hair, or breast development, sometimes before eight years of age (The Open University Sexuality Alliance, 2016; Blackburn, 2002). It was difficult from both the young adults' and parents' interviews for me to gauge whether precocious puberty was a common feature or not in this research, although the literature suggests that it is likely in this population (Craig and Lidstone, 2012; Sawyer et al, 2012).

Oak, another GP, described how her colleague dealt with contraception:

"One of my colleagues has dealt with an issue of contraception in the past, you know, around learning difficulties. That would be our remit, you know, and I would expect a GP perhaps not to fill out a mental capacity form but I would expect them to be aware of the law and I would expect them to refer [people] appropriately for contraception, when appropriate. I would probably expect them to use a family planning service and this is an issue for people with major life-limiting illnesses and learning difficulties." (*Oak, GP, CPF*)

The 'issue' raised by *Oak* related to her concern that contraceptive advice for people with disabilities should be provided in appropriate services and I suggest that she may not have considered a generic family planning service suitable. A key issue in the UK is that different rules and laws related to sex are usually based on age, mental capacity, the nature of the disability, and whether a disabled person lives at home or in a residential setting (De Than, 2014). This sometimes makes issues about sexual expression so complicated that many people don't understand what they can and cannot do and how to discuss contraception, (The Open University Sexuality Alliance, 2016). *Ellie* highlighted some of the particular issues facing GPS *Ellie*, in particular, she described the overall lack of opportunity that general practitioners had to care for young adults with LLTCs during their careers.

:

“In a GP’s [professional] lifetime, they [GPs] may only have one or two young people with life-limiting or threatening conditions on their books, and may never get really involved in them either because there are so many different services offered to these families and particularly the hospitals can control them an awful lot. So, in terms of hands-on GP interaction it is really rare I would say, so yes, no I mean in my previous job I happened to be the GP of one of our hospice young people but again you know actually I probably saw him more at the hospice than at, than in his own home and in primary care. I would absolutely expect that GPs would not feel skilled, experienced or empowered about the adolescence of people with life-limiting illnesses at all, let alone discussing the specifics of contraception, sex and all of that.” (Ellie, GP, CPF)

Similarly, Oak raised the importance of access specialist transition support:

“I suppose when I think about sexuality and some of the situations of transition and relationships, and menstruation, bodily changes and all that, I would actually feel very concerned about raising something in which I could not then offer them, the real and specialist transition help they [a person with a LLTC] need; I mean you would sort of naturally avoid that conversation.” (Oak, GP, CPF)

Both Ellie and Oak raised issues and concerns about the prevention of pregnancy and sex(uality). This echoes research findings more generally about disabled women’s reproductive and contraceptive rights (Earle and Letherby, 2002; Earle, 2001).

My interview with Oak (GP) provided a pertinent observation:

“I think anything to do with sexuality and reproduction is just not priority, but I think a sex life, if one could use that phrase, is not prioritised in a rationed health care system, so you know, if I am thinking about my older patients who have to fund their own Viagra, that is because their erectile dysfunction is not prioritised as much as their blood pressure. It is not perceived as such an important issue. I think there are probably some cultural judgements there about what is and isn’t the state’s responsibility to provide and so it is low on the agenda, sex and people with life-limiting illness.” (Oak, CPF, GP)

Oak and Elm’s discussions highlighted that GP consultations about puberty and sex(uality) with young people with LLTCs may not be considered part of their role but the remit of others, such as specialist palliative care doctors and nurses. Also, puberty in the context of the emerging sex(uality) of young adults with LLTCs is not considered a priority in the health arena. It is true that the lived experience, ambitions and values of adolescents living with such LLTCs *may* differ from those of healthy adolescents, and their physical lives relating to their expectations about life should include access to advice about puberty, sex(uality) and sexual functioning. The diagnosis makes people with LLTCs feel different (Kelly, 2013; Levi, 2006). Puberty and sexual functioning are important hurdles to overcome (Sawyer et al, 2014) and are also part of the uncertainty of living with a LLTC.

Summary

This first section has highlighted specific issues around puberty and how the young adults and parents managed these, particularly periods and erections. The presence of a LLTC during childhood or adolescence presents challenges (Kelly, 2013) such as hair loss and respiratory and muscle weakness associated with particular conditions (Kelly, 2012). Certain medications, as described by *Maserati*, may have negative effects on (male) sex drive, erections, ejaculation and orgasm (Wilson, 1991).

Hockey and James (1993, p.50) have argued that the individual's uncertain journey through life may have 'false starts, changes in direction and hidden obstacles' and the young people in this study were no exception. The transition from childhood to adulthood is seen as a 'normal' phase of the human life course for heteronormative individuals, which focuses primarily on the processes of identity and socialisation (Kelly, 2013). The experience for those with a LLTC differs; such as how their bodies differed or if periods or erections started earlier or later than for non-disabled people, and this leads to the next section: genetics and fertility.

7.2 *Genetics and fertility*

Two key aspects in reporting the findings in this section are: (1) the extent to which certain conditions were genetic, for example cystic fibrosis, and (2) whether participants considered they might be biologically infertile or childless, whether by personal choice. I was keen to understand the support and advice available to people with LLTCs who were or were not planning a pregnancy. I was particularly interested to listen to the views of young adults about genetics, the risks and the support which they or their supporters had been offered, and the extent to which genetic counselling affected participants' attitudes and thoughts about having children.

Some genetic disorders were apparent at birth (see Table 4), while others were diagnosed at different stages throughout childhood (see Table 4), and yet others manifested during adolescence and early adulthood (see Table 5). The issue of genetics was particularly important in this research as many LLTCs are genetic in origin, and sometimes without a specific name (see Appendix I). The subject of genetics was also relevant in order to help plan young adults' future lives, particularly in terms of their individual reproductive futures, , as well as for those who were likely to continue supporting them, such as partners (*Smidge, PF*, and *Dee, PM*) or parents (*Topaz and Liliana, MFs*).

Seven young adults, *Diamond, Lily, Vincent, Morris, Fiat, Mini* and *Austin*, two partners, *Smidge* and *Dee*, four parents, *Topaz, Liliana, Goldie* and *Pearl*, and five care practitioners, *Oak, Ellie, Peach* and the Transition and Youth Leads from *Sunshine Hospice*, all discussed genetic issues with me. I did not ask all the participants about genetics; some were shy or their health circumstances made it inappropriate at the time of my interviews, for example, *Jaguar, Lamborghini* and *Marmaduke* (YAs), and *Crystal* and *Emerald* (FMs). *Lily, Morris* and *Austin* (YAs) said that they had not been offered genetic screening. *Lily* would have liked to receive genetic screening:

"I think I'd like genetic screening. I have been trying to drop hints [to doctors], but I think I am going to have to out-and-out ask them [doctors]. But in terms of sexual health, I don't know if I would feel comfortable with them [the doctors]. I don't know why, it is just such a personal thing." (*Lily, YAF*)

Lily identified a gap in 'genetic testing' for her older sister as well as herself:

"Genetic testing? We can't, for example, test my sister to see if she has got the same gene as me which might be passed on to her children and equally as much because I haven't got a clue. I wouldn't know if I could pass on my condition, what the likelihood was of dominant or recessive or anything like that, I wouldn't know, because I haven't been genetically tested for it. They are not sure of the particular type of gene for the condition I've got. But the genetic side of it I think I could deal with and if I wanted to have children I could deal with all of that, but again talking about relationships and things like that, I don't know if I would feel comfortable." (*Lily, YAF*)

Here *Lily* raised several points. First, she recognised that she would "like genetic screening" and her need to request this from doctors. Second, that she would not feel comfortable discussing "a personal thing" such as "sexual health" with doctors, and

third, the availability of genetic screening for siblings. In Chapters 3 and 4, I noted that *Lily* had a rare, unnamed LLTC which was thought to be genetic in origin. Other research has highlighted that siblings may struggle to find accurate information about LLTCs and genetics (Brown, Coad and Franklin, 2017). There may also be implications for other family members (Brown, Coad and Franklin, 2017; Metcalfe et al, 2011).

At the time of my interview, *Lily* was just beginning to realise and understand the implications of genetics and the possibility of familial carrier status. This concurs with Metcalfe and her colleagues' 2011 research findings. In their research, they raised ethical questions about how much young people with LLTCs really understood about their condition and whether the timing of information and how it was explained was important.

When I explored why her sister had not been genetically tested, *Lily* replied that "she was not sure". *Lily's* mother, *Liliana*, spoke about the medical support and the genetic counselling the family had been offered:

"It was really through the internet and Lily doing a lot of research herself that we found out about different consultants that might be able to help [with genetic counselling] and have been absolutely fantastic, but we had a wonderful GP who was very supportive about genetic counselling and said I haven't got the time to look up on the internet, but she said whatever you feel, if you feel you want to go and see a consultant I'll write the referral letter. She said you're going to have to do the leg-work yourself, because she [the GP] was out of her depth on the genetics." (*Liliana, MF*)

Vincent, his fiancée, *Smidge*, and his mother, *Topaz*, described the genetic advice which both he, family members and his fiancée had accessed following *Vincent's* diagnosis of cystic fibrosis:

"I was around two when I was diagnosed [with CF]. Yes, I have an older sister who's thirty something, and she's a carrier. My second brother, he is not a carrier, he doesn't have anything apart from ADHD. He has been screened, but everyone [in my family] has yes, but he has this ADHD and then my youngest brother, he's a carrier. My parents are carriers. No one else [in the family] with CF that we know of obviously. I would imagine that part of my grandparents on either side, they haven't been tested. They will be carriers in order for my parents to be carriers." (Vincent, YAM)

Vincent had gathered quite a lot of information about genetics related to CF, carriers and how this might impact on other family members. Both Vincent and Topaz highlighted the importance of opportunities to meet with health or social care practitioners with whom they could discuss the implications of genetic risks as well as genetic testing; a finding echoed in other research with people with LLTCs (Brown, Coad and Franklin, 2017). At the time of interviews, Vincent was engaged to Smidge. Vincent had not yet received genetic testing in relation to his fertility:

"Obviously I have got to be tested, like a fertility test for myself which I haven't got round to yet. That's more of a sense of apprehension because I think I don't want to be, I know there's something I can do about it, but I don't want to be the one who lets the team down. I know that's a stupid thing to worry about, but obviously it's a very sort you know natural thing to worry about if everything's going fine, you don't want to be the last broken link you know. I know if I want to get to having children I have got to be tested, yes, so I will get round to that at some point." (Vincent, YAM)

Whilst Vincent was encouraging his fiancée to be genetically tested, it is surprising that he had not yet pursued fertility testing, even though he was planning to marry and have children. The literature recognises that many young adults confronting their

fertility or infertility may find such procedures difficult (Metcalf et al., 2011). Moreover, infertility and infertility treatments may lead to feelings of loss of control and compound such difficult feelings (Letherby, 2012).

Smidge had been screened for CF in hospital at both *Vincent's* and his family's suggestion as she and *Vincent* were planning to marry:

"Smidge was [genetically] screened not long ago [at the hospital]. She's not a carrier, well for all the common ones, all the ones they can test for, she's clear. Obviously, there's the ninety nine per cent chance, you know, like the one percent chance that she could be of a rare form, but yes she's not a carrier, so that's good." (Vincent, YAM)

Smidge was screened to ascertain the risk of her being a CF carrier. Most commonly, a couple will be concerned about the risk of recurrence of a medical condition that has already occurred within the family (Read and Donnai, 2012). Following genetic counselling, *Smidge* explained to me that she was not a CF carrier:

"It seems some time ago, March or April this year I attended [the genetic] counselling and screening [clinic at the hospital]. I got the letter with the all clear in May." (Smidge, PF)

Topaz, *Vincent's* mother, described some of the challenges which she had encountered whilst trying to access genetic counselling following a therapeutic abortion when it was thought that she may be expecting another baby with CF:

"But no real genetic counselling; nothing. I do think it was really, really bad. I did try to contact somebody once, but I know I actually never got to speak to anybody but I did try, because I wasn't coping with it [the therapeutic

termination] particularly. I thought I had, was coping with it, because in my head I thought well if I'm going to go forward with this I've got to get my mind set, I think I dealt with it okay really [the therapeutic termination]." (Topaz, PF)

Topaz had one child with CF, Vincent, and had had several miscarriages and a therapeutic abortion (see Table 6, Category 1). Being unable to have biological children challenges the normative expectations that all women are, or want to be, mothers (Earle et al, 2008). Mothers like Topaz who experience reproductive loss following a therapeutic abortion or miscarriage may find the subjects of termination and therapeutic abortions harder to discuss with care practitioners. This particularly impacted on Topaz who had previously undergone therapeutic abortions, but also the practitioners supporting people like Vincent, Topaz and other family members.

Topaz explained other family members' genetic history and how they had accessed genetic counselling and from where:

"I've got my eldest daughter, she's 35, she's a carrier, but she has three children. She didn't have any genetic screening until she actually got pregnant, and I sort of said well you need to look into it. And it [the results] came back that 'yes, she was a carrier' but she was already pregnant by that time. We're both carriers [Topaz and her husband] of the common one, the F508 [a specific mutation of Cystic Fibrosis transmembrane conductance regulator CFTR] which obviously Vincent is the double, he's obviously got the double fault. Yes, we know, we didn't know until we had Vincent diagnosed that we were even carriers. Yes, you have to both be a carrier to produce an affected child." (Topaz, FM)

Austin told me that he had not been offered genetic counselling:

“No, I don’t think there are hereditary links and we haven’t been offered genetic counselling. I’m not a whizz kid with the old family genetics tree at the moment.”
(Austin, YAM)

Whereas *Fiat* had ‘learnt’ about genetic screening over the internet:

“No, we never discussed genetic screening together. I learnt about genetic screening through Facebook and my friends on Facebook.” (Fiat, YAM)

In their research with 33 families who had members with a LLTC, Metcalfe et al (2011) observed that some parents encouraged and supported young people with discussions to make their own decisions about genetic testing. Similarly, some young adults in this research felt able to discuss and communicate their thoughts about their genetic condition with others, such as parents, sons or daughters (*Lily* and *Vincent*, YAs, and *Liliana* and *Topaz*, MFs). In contrast, some young adults did not discuss their thoughts and concerns about their condition with either their parents or clinicians (*Maserati*, *Morris* and *Austin*, YAs), and other researchers report similar experiences (Brown, Coad and Franklin, 2017; Metcalfe et al., 2011; Coad, 2008; Metcalfe et al., 2000).

For *Maserati*, ‘getting better’ was the focus of his attention and not specifically genetic counselling:

“I haven’t really thought about this yet [genetic counselling]. My focus at the moment is on getting better and living.” (Maserati, YAM)

Morris explained that his father had read about his illness but had not been offered genetic counselling:

“My father did a bit of reading about my illness. I think I may be the only person with this condition [ALD, see Appendix I], not sure. I am not able to have children because of my illness in case the child gets it. I have had scans on my head, brain scans, and lots of them you know. But no genetic counselling.” (Morris, YAM)

In Chapter 5, *Morris* explained that he couldn't:

“...talk about anything to do with sex and growing up and risks with my family, particularly with my mum, although dad's a bit better.” (Morris, YAM)

So whilst *Morris* could not have children because of his ALD, it seemed that he had not received authoritative information about genetic risks. Previous research has shown that neither discussing nor withholding information about genetic risks with young people until adulthood may impact on family relationships, the young adult's self-esteem (McConkie-Rosell, Heise and Spiridigliozzi, 2008; Fanos, Davis and Puck, 2001) and reproductive decision making (Fanos, Davis and Puck, 2001).

Dee said that:

“As we're not planning to have children, genetic counselling is probably not that important for us now. If we were, then that's a different story, with all Jane's complications such as diabetes and being blind.” (Dee, PM)

Although *Dee* acknowledged that he and *Jane* were not planning to have children, a couple might request specialist genetic counselling about possible reproductive risks and the options available to them, as *Vincent* and *Smidge* had sought. Most commonly, the couple will be concerned about the risk of recurrence of a medical condition that has already occurred within the family, such as cystic fibrosis in *Vincent's* family (Read and Donnai, 2011a, 2011b).

Parents mostly reported that they had not received information about the genetic risks, nor had received genetic counselling. *Daisy*, both as a parent and special needs educator, recognised that:

“Genetic counselling is important as well and we’re looking into that both as parents and as an educator [of a special education needs unit] and the risk to any offspring.” (Daisy, MF)

Similarly, *Pearl* and *Goldie* indicated that they would like to have the opportunity to speak to care practitioners about genetic counselling. At the time of my interview, *Pearl* had not been offered this opportunity:

“Well, I was saying to the Deputy Director of Care at the Hospice because Austin has got a brain tumour, that never throughout subsequent years in between did I feel that we needed much support, so I have never really looked for genetic counselling. We weren’t offered any [genetic counselling] but when he was 13, the operation, he had some recovery time, he went back to school, and you did part time, didn’t you, but nothing since. We learnt things from the brain tumour support group. I would go to the Hospice and Headway [for genetic counselling advice]. They’re easy to talk to.” (Pearl, FM)

Brown, Coad and Franklin (2017) argue that where communication about genetics is open, family members establish a shared understanding and reality that enables both parents and children to support each other about important genetic decisions, as well as seek help from genetic counsellors or care practitioners.

Goldie’s personal endeavours to learn more about her son’s condition were achieved by undertaking a palliative care course, (described in Chapter 5):

“Anyway, I know my own son. I waited until I finished my own child care course. I came out with A stars. Again, I was told my son [not interviewed] had dyspraxia see Appendix A} he kept falling down the stairs. I went to the doctors ‘and things then started to move. And because of my training, I learnt more about my son’s condition”. (Goldie, MF)

Five care practitioners discussed genetic counselling (*Walnut, Pine, Oak and Ellie, GPs, and Peach, CPF*). Their interviews focused on the dearth of genetic counselling specialist training, genetic counselling, and the distances required to access services. *Walnut* and *Pine* talked about the paucity of local genetic counselling facilities and noted:

“Genetic counselling? Hmm. Here there are real gaps. Not enough counsellors, screening programmes, specialist staff or facilities locally. [The service] It’s pretty thin, long waiting times and that’s a real worry today.” (Walnut, CPF)

“We did a lot of preconception counselling, she was referred to a genetic counsellor, [she is a young woman with a LLTC who was not a participant in this research]. We took advice, we dealt with her medications and that was my job. So for those kind of medical things around it, contraception, genetics, even some of the basics of it, you know lubrication, that sort of, I think when it’s medical, I think that is my role and it might even be my role to raise it, so I would raise the subject of contraception and genetics for example with my patients.” (Oak, CPF, GP)

I suggest that *Oak* felt it was within her remit as a GP to discuss preconception counselling and contraceptive advice with people with LLTCs. *Peach*, a medical student, appeared to understand the meaning of genetic counselling, but at the time of interview, she had not received much training on this subject:

“So individuals diagnosed with a genetic condition, that’s then you’d go on then to do genetic counselling and family and things like that, but that’s it. The actual process of what [genetic counselling] entails, we haven’t received much training yet on this.” (Peach, CPF, Medical Student)

Ellie, a GP, described a different experience of her medical training:

“Genetics: yes, definitely. I mean as an undergraduate genetics medical student because obviously medicine figures in genetic DNA, so definitely genetics we learnt a lot about, not necessarily just the counselling but the whole process of it and then I would say certainly genetic counselling has become a bigger topic throughout my postgraduate years in that its more widely recognised, more widely requested now and more widely available I think than it was when I started out for sure.” (Ellie, GPF)

Oak then talked about the various services, hospices and palliative care provisions that she might draw upon for genetic counselling:

“Well it depends, you know. I live in a county that has the Hospice so I would refer them to them (the young adults for genetic counselling). In the absence of the Hospice, it depends how old they were, if they were still under paediatric palliative care service I would use that, I would. It depends what the problem was. If there was a contraceptive issue or genetics issue, I’d refer appropriately, you know, so I think yes I would do that. I don’t really have any other, so I have a GUO [Genito-urinary outpatient] service, a gynae service, a paediatric service and a palliative care adult service but I have never had a conversation or neither am I aware of any service through there that would be particularly around sexual health. They do have some counselling at the Hospice for perhaps if somebody

had a psychosexual problem but dealing with specific psychosexual health is very, very hard.” (Oak, GPF)

Summary

Overall, this section has presented a mixed picture of whether the different participants were able to access genetic counselling, from whom, when and where. Young adults and parents valued being able to discuss genetic conditions affecting their family, with care practitioners in hospices or with genetic counsellors. In cases where young adults had not always received explanations from their parents or care practitioners about their genetic condition, this was stressful for young adults, parents and partners, and on their relationships with each other and with their wider social networks (Metcalf et al, 2011). Once family communication becomes more open, creates a shared reality that enables parents and young adults to support and help each other in discussions (Brown, Coad and Franklin, 2017), such as, reproductive opportunities, choices and practicalities, which are the focus of the next section.

7.3 Reproductive opportunities and the future

In Table 6, I set out a typology of reproductive loss organised under four categories, the first three of which are adapted from Graham et al (2012) and Earle et al (2007) and address the different types of reproductive loss and whether they are medically or socially constructed. I have included and adapted a fourth category from these research findings, and considers the particular types of reproductive loss of relevance to the participants in this research. I have also included the participants where the first three categories also apply.

Most young adults in this research said that they were not planning to have children (Jane, Lily, Marmaduke, Fiat, Morris, Jaguar, Lamborghini and Mini, YAs). Notable

exceptions were *Diamond* (YA) whom at the time of interview was married and had had two children. *Vincent* and his partner, *Smidge*, were engaged to be married. I foreground this section by addressing the findings from the couples who were in established relationships: *Diamond, Jane* and *Vincent* (YAs) and their partners, *Smidge* and *Dee*. (*Diamond's* husband declined to participate in this research).

Diamond had received treatment for her childhood leukaemia over a period of time, and subsequently experienced dysmenorrhoea as well as regular and heavy periods during her transition into adulthood. Her potential 'infertility' was a concern for her, because of the presence of 'polycystic ovaries' (see Appendix A), which may cause infertility problems in some women. *Diamond* also had childhood leukaemia and she thought that there may have been a link with her father who had died from a 'blood' cancer in his sixties:

"I didn't know if there would be any problems or issues in me regarding pregnancy as a result of my Dad's blood cancer, leukaemia or my own leukaemia." (*Diamond, YPF*)

Diamond had always wanted to have children but had been advised by oncologists and infertility specialists that this would be unlikely because of her life-threatening condition. *Diamond* subsequently married and gave birth to two able-bodied children. She described the challenges that she had encountered to have children. Regardless, *Diamond* had sought help from an infertility clinic and stated:

"Well, I'd been married for five years before I had my first daughter. So we'd been trying for about a year. And of course, we didn't believe it, so I had to take lots more [pregnancy] tests and they were all positive. So we [she and her husband, who did not take part in this research] were quite ecstatic and delighted and then went to the appointment and told him [her husband] with

delight that I was pregnant so he discharged me from the fertility clinic. I was a success story.” (Diamond, YPF)

Both of *Diamond’s* pregnancies progressed satisfactorily and she gave birth normally to both her children. *Diamond* explained that clinicians were concerned that she might encounter difficulties during her labour and she described this:

“I was induced with my first labour because my blood pressure had gone up a bit and there was some protein in my urine, that sort of thing, but no huge concern, it was just a bit of a ‘well we’ll do it just in case there are any complications’.”
(Diamond, YPF)

However, *Diamond* also stated that clinicians were concerned about her life-threatening condition and that:

“I might not cope with the labour because they thought there might have been something underlying that the treatment had caused that may cause problems with the baby, placenta. Not just with the impact of labour and the stress that it places on your body and I might not be strong enough really for that.” (Diamond, YPF)

Diamond also acknowledged that her pregnancies were monitored more closely on account of her childhood cancer:

“They [the doctors] wanted to check, they did an echo to check that my heart was okay and I wasn’t going to have any problems and I also had some respiratory function tests and they told me that they wanted to have that each time I was pregnant just to check.” (Diamond, YPF)

Following her childhood cancer, *Diamond* told me that she had developed several health issues which she attributed to her childhood leukaemia:

"I had also suffered with endometriosis. My womb thickened, which is one of the other things that came from the laparoscopy. Had a second laparoscopy after my second child and they said that the endometriosis had deteriorated, there was more areas affected so actually when my youngest daughter was 18 months, just over a year old, I had a full hysterectomy. I'd also had in-between, pre-cancerous cells on a smear test so I had to have a colposcopy for that and so because of all the issues together and the pains and everything; they did a full hysterectomy when I was 36 with removal of ovaries and cervix as well. So yeah. I've had no periods since then... which is great! And I didn't recover very well either. I was quite poorly afterwards and I thought at the time, 'oh well it's the worst thing I've ever done'. But actually, years on, it was probably the best thing I've ever done. It was very difficult at the time. I've had cardiac ablations for years; the pathways were going a little bit haywire. So that affected me during the pregnancies as well, as when I wasn't pregnant." (Diamond, YPF)

Diamond reported that her first daughter, who was nine years at the time of my interview with her, reported similar signs and symptoms to those of her own childhood leukaemia:

"I took Amber [pseudonym] to the doctor. I was worried. I don't want her to get childhood leukaemia as well. She seemed to be presenting similar signs as me at the same age. Such a worry for us all." (Diamond, YPF)

Vincent, at the time of interview, was planning to marry *Smidge*, who did not have a LLTC:

“Smidge and I would love to have children. We’ve both had genetic counselling, and will probably need IVF and we’re considering this if my body can cope with this but as I said earlier I haven’t got round yet to the fertility testing.” (Vincent, YAM)

Jane, who was in a steady relationship with Dee, stated that she did not wish to have children on account of her rare and complex conditions:

“And we spoke about having children and Dee spoke to me about this. He said because I’ve got a life-threatening illness anyway I said to him, look, are we going to have children together? He said, look, I’d love to have children with you but I’d rather keep you. And he doesn’t want to put me in more danger because giving birth could cause me, you know, problems.” (Jane, YAF)

Dee told me that he and Jane were not planning to have children and that there was also a risk of pregnancy and unplanned children:

“If we did get to the point we decided to think about children, I think it would be a long sit down with a specialist somewhere because of the possibilities of what it could do to her health and the child. We wouldn’t want an unplanned pregnancy.” (Dee, PM)

Other young adults discussed reproduction and children with me, for example, Lily (YP), Liliana (MF), Austin (YP) and his mother, and Marmaduke’s mother:

“I don’t think my body could cope with having a child but I would be happy to adopt if I was well enough to be able to look after a child but I have always felt like that actually. I said to my mum when I was about 11, I don’t want you to be upset, and my mum was panicking at this point, and I said but I don’t want to

have children, I said if I want to I will adopt, I said but I don't think I want children." (Lily, YAF)

Lily also discussed the feasibility of adoption:

"I think again that probably reflected whether I wanted children, because I would either have to have a sperm donor if I wanted a biological child or I would just adopt, and I always thought, what is the point in the chance that I am going to bring a disabled child into this world? I would have been quite happy with that, and I don't feel that need to have my own child, that is biologically related to me, because I could love a child whether it was or wasn't. Again, I don't know that was because I was starting to get ill and I kind of knew that there was something going on or whether that was just a choice I made." (Lily, YAF)

Parents recognised that they were likely to 'be responsible for bringing up the child, if their son or daughter died' (Liliana, Topaz, MFs):

"Rather than worry about what could happen let's just do it as it happens [see if Vincent and Smidge can have a child]. So, that's the way they're going I think, they're just going to let it happen. But of course, I worry about our own health as Vincent gets older. What happens if Vincent dies? Smidge and us, we'll have to raise his child." (Topaz, MF)

Here Topaz acknowledged that the presence of children (grandchildren) might coincide with the young adult's parents' deteriorating health, thus possibly leaving responsibilities to others to care for and bring up a (grand) child. There is an emerging literature about older parents supporting surviving adults with LLTCs (Carpenter, 2012) but at the time of writing there is an absence of research on the implications of

partners or parents supporting young adults with LLTCs who have had their own children.

Several care practitioners discussed their views about young people potentially having a child (*Primrose, Oak, Ellie and Daisy*). Their responses varied. *Primrose*, a nurse at *Sunshine Hospice*, had a disabled child and spoke about people with LLTCs wishing to parent:

“Good luck to them if they want to parent... as long as they don’t make a decision willy-nilly.... A 32-year-old in Scotland who is having a baby... will choose an embryo who is not a carrier of the condition... who are we to judge who brings up a child... should he be allowed to bring a child into this world... if that is what they want to do... fine, but they and services need to remind themselves that inevitably they [the young person with the LLTC] will die. The legacy will be in the child who is going to be born... Good on yah.... I would back them... for goodness sake... life is too short... But this is a sad place... but some staff will have different views. I know that I shall go to my maker knowing that I have done a good job... and a happy person.... I had a disabled child... you do your best... you grab life with one hand. Who is to know... what will happen to us all... some staff have iron rods up their bums and need to change their attitudes towards the sexuality of cognisant young people and then we would all be in a better place.” (Primrose, CPF)

Summary

In heteronormative relationships, life is expected to progress in an orderly fashion. This was not the case for the participants in this section, whose experiences were complicated by the presence of their LLTCs. For these young adults, the uncertain life course, compounded by uncertainty around fertility and reproduction, interrupted or

curtailed that desired trajectory, as was evidenced by *Vincent (YA)* and his partner, *Smidge*.

7.4 Discussion

The findings have highlighted several important points. These include the importance of addressing the practicalities of puberty, genetics, issues of reproduction, and the impact on young people and families should young adults want to ‘have a child in future’ (*Vincent, YAM*), as well as the ethical and legal considerations of this. Sexual and reproductive health and access to information are important features of health, However, people with LLTCs were not always provided with the advice and information they required to appropriately plan. For non-disabled people, this topic is addressed in schools, at home, in doctors’ offices, or in the care of other human service professionals. People with LLTCs in this research were not always asked about their sexual or reproductive needs, as society assumes that they cannot or do not have sex (Liddiard, 2011).

Puberty and adolescence

Garbutt (2010) and Hollomotz (2010) argue that the denial of even the basic knowledge and information addressing puberty and reproduction for disabled young people remains tied to dominant notions of infantilisation (Liddiard, 2011). Often young adults are assumed to be:

“incapable of forming substantial life preferences, learning the skills necessary to negotiate sexual choices, or making meaningful decisions in general.”
(Wilkerson, 2002, p.43).

This was particularly important for *Jane*, in terms of her managing her menstruation, contraception and reducing the risk of pregnancy. Treatments experienced by cancer survivors such as *Diamond* and *Maserati* included radiotherapy and chemotherapy. The use of intensive treatments may make people with LLTCs feel that their body is no longer theirs, no longer 'normal', and may result in an altered sense of body image and an inability to confidently enter intimate relationships.

Genetics

There are currently between 4,000 and 6,000 diagnosed genetic disorders (Rare Diseases UK, 2018). This is extending as genetic testing and identification of genetic disorders improves. Many cancers are now known to have a genetic component. It is estimated that one in 25 children is affected by a genetic disorder and that 30,000 babies and children are newly diagnosed in the UK each year (Rare Diseases, UK, 2018).

Genetic Counselling

'is the process by which individuals or relatives at risk of an inherited condition receive advice on the consequences and nature of the genetic disorder, the probability of developing or transmitting it, and the options open to them in management and family planning.' (NHS website, online)

Participants wanted to access genetic counselling. The research findings revealed several reasons for this. Although access to genetic counselling was recognised as a barrier (*Vincent, Smidge and Topaz*), the participants were particularly keen to understand more about the genetic implications of their LLTC and how these might impact on other family members (*Lily, YAF and Liliana, FM*) as well as the risk to future offspring (*Vincent, YAM and Smidge, YAF*).

These research findings have highlighted issues that related to participants' explanations of inherited genetic risk to their children, the importance of sharing information, the young adults' understanding of explanations that they may or may not have received, the emotions evoked for family members about puberty, genetics and reproduction, and the support and guidance received from health professionals. Providing information, checking understanding, and explaining and managing the emotional feelings that arise were integral to supporting families coping with the risks of a genetic condition. However, I would argue that some young adults and parents struggled with one or more of these components and required more support specific to the stage and family members' transition of readjustment to the impact of the genetic condition.

It is not an unreasonable expectation that most human beings wish to pass on something of themselves to the next generation, usually in the form of their genetic inheritance. Two young adults in this doctoral research shared these views (*Diamond and Vincent, YAs*). The absence of being able to achieve this may cause emotional strain and impact on individuals with a LLTC, such as loss of self-esteem and depression (Kelly, 2013). Infertility, which may have many causes, may present differently depending on the individual. Until recently, little has been documented on how parents explain to their children the risk of inheriting a gene that may cause disease in the young person or their future progeny.

The findings have highlighted how genetic-risk information is accessed and communicated between family members and care practitioners, and why this is important. Communication about genetic risks and the availability of genetic counselling services were both recognised as important by all participants. Of equal importance were the views and experiences of families about conditions that may vary in inheritance pattern, age of onset, rarity, morbidity and life expectancy (Metcalf et al., 2011). As Metcalf and her colleagues (2011) noted, the detailed content of family

discussions in their research varied between the genetic conditions and their presentations. The young adults expressed that know about genetics would help them to come to terms with the risks, either to themselves or to other family members, including their own future offspring (*Diamond and Lily YAFs*).

Fertility and reproductive opportunities

In this research, only *Diamond* had given birth to two children following diagnosis and treatment of her childhood LTC. The other participants were interested to learn more about their reproductive opportunities and choices, facilities available, as well as the risks (*Vincent and Smidge, YAS and Topaz, Liliana PFs*). Research on the long-term effects of fertility on sexuality and intimate relationships has largely focused on young adult survivors of childhood cancer (Kelly, 2013; Crawshaw et al, 2009; Zebrack Stevens et al, 1998). *Diamond's* findings have highlighted that the potential or actual loss of fertility may become more important as the young adult begins to contemplate long-term relationships and parenthood, as set out in Table 6. As other research confirms; reduced fertility is a common feature associated with cancer diagnoses as well as the short- and long-term effects of treatments (Kelly, 2013).

Until recently, sex(uality) has not been seen as an integral part of the lives or lived experiences of disabled people in modern Western societies (Liddiard, 2018).

Moreover:

'Disabled people are expected neither to reproduce nor be reproduced.' (Earle, 2001, p. 435)

Young adults with LLTCs, particularly those with cystic fibrosis, like *Vincent*, may now approach developmental milestones, including sexual and reproductive ones, at a similar time to their non-disabled peers. This was previously not the case (Frayman

and Sawyer, 2015; Edenborough and Morton, 2010). Yet, for many people with LLTCs, sexual and reproductive health (SRH) may be profoundly affected by their condition(s). The young adults reproductive aspirations and futures influenced not only by the uncertainty of their life course, but genetic and reproductive implications (Frayman and Sawyer, 2015). Arguably this creates another transition point in the young adult's life, the point where the young adult may or may not be considering their reproductive choices and the obstacles and challenges that this may present to them in future.

Reproductive rights have been central to social science literature, particularly the rights of women to retain 'control of their own bodies' (Tilley et al, 2012;). The experiences of disabled women are often addressed in the context of reproductive technologies (Shaw, 2004). It is only recently that discussions such as those between *Jane* and *Walnut* about Depo-Provera have been included in mainstream).

Professor Hawking was diagnosed with a rare form of motor neurone disease (ALS) when he was 21 years old and died in 2018, see Appendix A. Sex may still be manageable with this condition. Sexual function is not usually affected, but sexual expression may become more difficult if mobility and movement are. Stephen Hawking fathered three children with his non-disabled wife.

7.5 Conclusion

In this chapter, the subject of reproduction was important to some, if not all, young people with LLTCS and their supporters. Although most young adults in this research stated that they did not wish to have children, young adults stated that they wanted to retain the choices and aspirations to determine their own reproductive journey (*Diamond* and *Vincent*, YAs, and *Lily*, YA). In addition to the complexity of the LLTC, the young adult participants often fell victim to prejudices and taboos about reproduction. The participants would have liked to receive more information about the genetic risks

and the implications for reproduction. The challenge for young adults and their supporters is learning to recognise the uncertainty of their LLTCs as 'an emerging adult'. Finding a balance between the demands of their illness and their hopes for their reproductive future, while recognising and enjoying relationships and accomplishments were important to participants. The findings have highlighted a number of implications for policy and practice: the need to recognise that people with LLTCs consider puberty, genetics and reproduction important discussion topics and not to ignore the constraints experienced by individuals who were expected to die young, but are now reaching adulthood.

Chapter 8

Conclusions and recommendations

Introduction

After a complex and wide-ranging research effort, drawing mainly on an interpretivist, qualitative approach, I begin my concluding chapter with a (re)statement of how the theoretical perspectives in Chapter 2 relate to and underpin my data findings in Chapters 4 to 7. At the heart of this research were 13 young adults with LLTCs, their sex, intimacy, relationships and reproductive choices. These were explored in a series of in-depth, face-to-face, semi structured interviews. In order to give added depth and alternate perspectives, I also sought the views and contributions of family and professional supporters in interviews with two partners, ten parents and ten care practitioners.

A life course perspective has provided an important theoretical framework through which the views and experiences of young adults with LLTCs, particularly in relation to their sex(uality), have helped make sense of the data. In chapter 2, I presented the literature related to the life course perspective, drawing primarily on the seminal works of, for example, (Elder, 2000, 1985, 1978), biographical disruption (Bury, 1991, 1982), liminality (Turner, 1974, 1969), transition theories (Van Gennep, [1909] 1960), and uncertainty (Peou and Zinn, 2013; Charmaz, 2004;1999). These concepts represent different but compatible perspectives and worked well in elucidating the different research questions and findings in this research. Rather than adopting one perspective, I moved between them to form the bigger picture within the life course in my presentation and discussion of the research findings, see Chapters 4 to 7. This was a two-way process; my findings confirmed and illuminated the theories, and the theories helped to elucidate and make sense of my findings.

8.1 *Aspects of life course*

Transition, liminality, and emergence

Palliative care services emerged fifty years ago as an adult-based service and subsequently expanded to include services for children and young people, childhood services (see Section 1.7). These research findings have looked beyond the established adult and childhood services to focus on the emerging group of young adults with LLTCs; a growing population, at a new transition point in their lives, between adult and childhood services. I found that this third and distinct group of young adults required services beyond the reach and scope of either of these established service groups. These young adults were transitioning from adolescence to adulthood, from wellness to illness, and they not only sought health and social care services and advice appropriate to their needs, but wanted to be autonomous, albeit that total independence from carers was not always feasible. They wanted to socialise, to talk about and experience sex(uality), all in the shadow of their profound and often simultaneous, deteriorating health (*Lucy YAF and Vincent YAM*).

In essence, these young adults present a new dimension to holistic service requirements, as yet unprovided for, where adolescents are currently in a liminal state, falling 'betwixt and between' adults and children's services, requiring specialist support and services, including the recognition and acknowledgement of their sex(uality). Hitherto researchers have failed to adequately classify and report on young adults with LLTCs, and this has been for two reasons; first, because they were not never expected to reach adulthood. Second, having reached adulthood in terms of chronological age, they still have an uncertain and shortened life expectancy within the context of a normative life course trajectory (Earle and Blackburn, under review). As these young adults were not expected to survive beyond the transition from childhood to teenager, from teenager to adulthood, their specialist care was not

previously a consideration as most young people would not have survived long enough to access sex and relationship education (SRE), let alone explore and consider their reproductive options and the associated genetic risks (*Vincent YAM* and *Smidge, PF, Topaz, MF*). Liminality problematised the boundaries and intertwined nature between the biographical disruption and the pivotal transition points along the uncertain life course for all the participants' in this research.

Uncertainty and transition and turning points

The illness trajectories for the young adults with LLTCs have highlighted the fact that they have differing social, emotional, physical and intimate requirements. Uncertainty as to when they might die adds a degree of complexity to their care, and the particular individual support which they require during their transition into adulthood (www.togetherforshortlives.org.uk) which may be limited but should not be limiting.

There was wide variation in the service provision and priorities participants experienced, and variation in their rights to access formal, face-to-face advice about sex(uality). Several participants (*Austin, Morris, Maserati, Fiat, YAMs*) said that this was the first time in their lives they had spoken about sex(uality), and sometimes it had a profound impact upon them (*Lily, Morris and Vincent, YAs*). This was a turning point; for the first time, they had 'permission' to share and discuss their sex(uality) with someone, albeit a researcher.

For over half a century, the right to sexual experiences and fulfilment have been central to the politicising of sexuality for marginalised groups, particularly in relation to people with disabilities. Participants in this research found it difficult but wanted to talk about sex and relationships. This subject was considered personal, private and taboo (*Jane, Maserati, Fiat, and Mini YAs*). It was made more difficult because for most individuals, although their life course trajectory and expectancy were uncertain,

their lives should not be limited but lived to the full (*Lily, Jane, YAFs* and *Liliana, Topaz, PFs*). All the young adults were living with either physical and/or sensory impairments (see Table 2, Chapter 3), as well as experiencing periods of acute and chronic illness and treatments. Unfortunately, a lack of co-ordination between adult and children's services meant that these young adults fell through the gaps and frequently missed out on vital care and advice, particularly in relation to their sex(uality) (*Maserati, Vincent, YAMs, Diamond, YAF* and *Topaz PF*).

The need for intimacy, love and sex, which was not exploitative or solicited via the internet, often went unrecognised by parents or health and social care practitioners. Yet social media was acknowledged as accessible and important to obtain information and to communicate with friends in the absence of face-to-face dialogue (*Jane, Austin* and *Morris, YAs*).

Addressing potential dying and death across different transition points throughout the life course created deep uncertainty for all participants, but particularly the young adults and parents in this research. Maintaining a boundary between 'life' and 'death' was important but was sometimes influenced or controlled by parents and care practitioners, even when young people tried to resist this during their emerging adulthood (*Morris* and *Lamborghini, YAMs*).

Uncertainty and biographical disruption

Several young adults in this research felt they were living on 'borrowed time' (*Lily, YAF* and *Vincent, YAM*). They experienced progressive and deteriorating conditions that often impacted on their health and social wellbeing. Their emerging adolescence often coincided with their deteriorating physical conditions and health (*Jane, YAF* and *Maserati, YAM*). Sometimes the young adult and their parents received initial confirmation about their condition but this was often without a label because their

LLC was rare (*Lily, Jaguar, Fiat, Lamborghini and Marmaduke, YAs*) and participants' were uncertain as to their prognosis.

Most of the young adults were still living with family and had experienced crises and discontinuities in their lives, leading to long periods in hospitals or in hospices, which disrupted their lives (*Lily, Marmaduke, YAFs, Morris, Maserati, Vincent, YAMs*). Many of them were reliant on carers and were less likely to live independently in comparison to non-disabled people of a similar age (*Lily, YAF*). For some there was a shift towards life-long dependence on others (*Jane, Marmaduke and Vincent YAs*), notably to partners, families or carers (*Dee, Vincent, Ps; Goldie, Precious, Topaz and Liliana, MFs, Silver CPF*), either at home or in residential settings.

These factors created barriers or disrupted opportunities for social participation that might enable them to develop relationships. This became more challenging once they left school or adult education, because the young adults were spending more time at home with family **and** they had fewer opportunities to socialise with their peers (*Marmaduke, Fiat and Mini, YAS*).

The lived experience of 'borrowed time' created uncertainty, disruption and barriers for young adults and their supporters particularly in relation to talking about sex. Young adults lived with concerns about potential side effects of treatment, the fear of a condition's return or its exacerbation. There were also those who had survived a life-threatening illness (*Diamond, YAF*) and/ or the emotional changes of living with a LLTC (*Maserati and Morris, YAMs*), or those whose health was in an on-going decline (*Lily, Jane, Maserati and Vincent, YAs*). Living with a LLTC altered the meanings of the participants everyday encounters and these changed significantly with the onset or recurrence or deterioration of their illness, as specific aspects of the condition presented over time and not necessarily within a defined period (*Diamond, Jane and Lily, YAFs*).

Pivotal moment of diagnosis and its impact on sexuality

The young adults' responses to my 'ice breaking' question about their LLTC, partitioned them in a meaningful way (see Chapter 4.1). Their LLTC was central to understanding the lived experiences of their illness and how this impacted on their confidence to talk about sex. Both young adults and parents talked about the diagnosis, noting particularly if it had been explained to them, when and how it occurred, if it was confirmed or not, or if it were so rare that it remained without a label (Rare Diseases UK, 2018). These findings suggested that the point at which the young adult received their diagnosis impacted on how that individual addressed or acknowledged their emerging sex(uality) and created uncertainties. For *Lily* and *Maserati*, YAs, who were non-disabled and purportedly without a LLTC until their teens, their emerging adulthood experiences differed from those who had grown up from childhood, recognising that the presence of a LLTC may have implications for their physical and sexual development (*Fiat, Jaguar, Vincent and Lamborghini, YAMs*).

This research has built upon the idea that there can be 'innovation' within the typical life course, brought about when the temporal ordering of life events, complicated by the presence of a LLTC, departs from previous norms and becomes routinised, restricted, and almost quasi-institutionalised and where events and roles may change over time. The emergence of this new population of young adults with LLTCs can be recognised as an innovation within the life course and thus a life course perspective was ideally suited to explore the views about sex(uality) in this emergent population (Earle and Blackburn, under review). The next section focuses on the key findings from this research.

8.2 *Research questions: key findings*

In this section, I focus on the key findings from the research, in particular, exploring the extent to which I have been able to answer the four research questions which I set out to explore, (see Chapter 3). I presented and discussed these in Chapters 4 to 7, locating them in the wider contexts of young adults with LLTCs and the existing knowledge of disabled sexualities. Question 4 in particular focuses on the parents and care practitioners findings.

Question 1: What are the views and experiences of young adults with LLTCs about relationships, intimacy, sex and reproduction?

These findings were mainly discussed in Chapters 6 and 7. For young adult participants, their opportunities and experiences of sexual and intimate relationships were mediated by the lived experience of their LLTC. For the most part, the LLTC was a primary focus through which they conceptualised the rest of their lives. All the young adults stated that they were heterosexual, although two people stated that at times they felt 'sexually ambivalent' (*Maserati and Austin, YAMs*), one of whom questioned asexuality. Furthermore, the 'hard physical realities' (Wendell, 1996, p. 45) of the LLTC were significant to participants and, for most young adults, impacted upon their ability to engage in intimacy or sexual relationships.

Both the young adults and parent participants were frequently reminded throughout their lives that the young adult was likely to die before adulthood, and consequently learning about and engaging in relationships, intimacy and sex 'were not a priority' (*Pearl, FM*). When a young adult lives in the shadow of death but is desperate to live, that person often has to assert his/herself in order to engage with life). Most non-disabled young adults are unlikely to think about death on a daily basis, particularly if they have no apparent reason to do so, or do not have a significant illness which may

limit and impact on their everyday lives. Enjoyment, socialising, experiencing love and intimacy are acceptable 'norms' in the lives of non-disabled people. This is often based on the confidence that life will go on and not be interrupted in the way experienced by people with LLTCs (Beresford, 2004). Where death is potentially lingering, the concept of death for people with LLTCs may be an approaching reality that may lurk in the background, even when there is a desire to 'live life to the full' (Watts, 2018)

Relationship experiences

Five young adults had had a girl or boyfriend, and three stated that they had had an intimate relationship (had sex). One young adult had had children (*Diamond*); another couple (*Vincent* and *Smidge*) were planning to marry, hoped to have a child and were exploring *in vitro* fertilisation (IVF) programmes. One couple (*Jane* and *Dee*) stated that they did not want to have children because of the potential risks of having a child with a LLTC. The majority of the 13 young adults had not 'had sex' or been physically intimate with the opposite or same sex. Given the numbers and different LLTCs considered, I am unsurprised by these findings. Whilst it was positive to see a small number of participants embarking or considering relationships and parenthood, it was disappointing that my findings concurred with Liddiard's sexuality research findings, where disabled people for the most part seem unable to experience sexual agency and engage in relationships because of the limitations of their LLTC, because they are

'...presumed to lack the capabilities and capacities to embody and experience sexuality and desire, as well as the agency to love and be loved by others and build their own families, if they so choose.' (Liddiard, 2014, p. 215)

All groups of participants recognised the importance of companionship with others, particularly the young adults' opportunities to engage with their peers. Most groups of

participants recognised that sexuality should be addressed as a right and not enmeshed in a disease (Taylor, 2012).

Young adults with LLTCs appeared to experience different levels of physical and emotional maturity, as well as diverse life experiences (Frayman and Sawyer, 2015).

Reproduction

All groups of participants discussed reproduction; their hopes, fears, aspirations and considerations for the young adults. Most young adults had not (yet) considered having children. For *Diamond*, having two children was a positive experience, although shrouded with concerns about cancer presenting in her own children.

When a person's life is considered to be limited, that person might have a greater need to leave a legacy, and this might be a desire to have children. Naturally such choices have to be balanced against the risk of passing on the specific LLTC to a child, but the desire to have a child may not necessarily be wrong nor indeed be suppressed (Tilley et al, 2012).

Moreover, the process and consideration of IVF was recognised by *Vincent*, *Smidge* and *Topaz* to be emotionally and physically challenging as this was considered a form of reproductive loss if assisted conception was likely to fail (Graham et al, 2012). There may be important considerations as to who raises the child if the young adult dies following the birth of their child; as partners, parents or carers may begin to experience ill health themselves in their own advancing years (Mitchell et al, 2016).

All groups of participants felt access to genetic screening and counselling was important and should be provided earlier. Four young adults (*Lily*, *Jane*, *Austin* and *Maserati*) said they would have liked to receive genetic advice and counselling. One

young couple (*Vincent* and *Smidge*) and *Topaz (FM)* had received genetic counselling but would have liked to have received information and advice much earlier.

Parents who had had a child with a LLTC felt that genetic counselling should be accessible to other family members and that this had not always been the case. Opportunities for parenting and options relating to contraception, assisted fertility and obstetric care are expanding as people with LLTCs are living longer, but these also present different ethical considerations (Frayman and Sawyer, 2015).

Care practitioners highlighted a dearth of local genetic counselling facilities. The two doctors (*Oak* and *Elm*) questioned who was the best person or service to provide genetic counselling and overall felt that their own training and expertise were inadequate in this arena. They thought that there was a role for specialist genetic counsellors.

All groups of participants stated that relationships and intimacy, love, sex and reproduction were important. For young adults, the meanings and interpretations of relationships, intimacy and sex differed, and they often felt very shy discussing such topics. Sex was not just about 'having sex' and penetration; other features, such as touch, smell, kissing and cuddles were equally important (Blackburn, 2002). Most young adults felt the physical limitations of their LLTC were a barrier to even attempting 'to have sex' (*Fiat* and *Lily*, YAs). However, many young adults would love to have a partner and experience a relationship.

Several parents found it difficult to discuss sexual matters with their sons and daughters. Where care practitioners had received specialist sexuality training about supporting people with LLTCs, they felt more confident in discussing sexuality and relationship matters (The Open University Sexuality Alliance with Together for Short Lives, 2016). Frayman and Sawyer (2015) note that young people with cystic fibrosis,

and their parents and clinicians, acknowledge barriers to such discussions, including embarrassment, the importance of privacy, time, difficulties in initiating discussions about sex because concerns about other issues related to the LLTC were prioritised.

Question 2: To what extent are young adults, over 16, with LLTCs prepared through education and information, and enabled to make relationships, and sexual and reproductive choices?

This question was mainly addressed in Chapters 5 and 7 and explored preferred options about sex and relationship education (SRE) with all groups of participants. Overall, most young adult participants had received formal, generic sex education at school or college, but not specifically about the sexuality of people with LLTCs. SRE was mainly designed for non-disabled people. Most young adults could access some forms of SRE information online but stated that they required earlier and on-going access to both specialist SRE, and both formal and informal SRE. They thought it should be ongoing and available throughout life. Organisations such as the Sex Education Forum UK, the Family Planning Association (FPA), Brook Advisory and the Sexual Health and Disability Alliance (SHADA) have lobbied successive governments to provide statutory SRE as part of a young person's right to receive statutory personal, social, health and economic education.

As noted in **Question 1**, all groups of participants felt information about genetic screening/ counselling was important and that this should be accessible earlier, but was not.

GPs and nurse practitioners stated that specialist sexuality training related to palliative care and, more specifically LLTCs, should be part of their undergraduate and postgraduate specialist palliative care training, but was notably absent at the time of interview.

Question 3: What are the relationship opportunities and experiences for young adults with LLTCs?

The young adults acknowledged that the restrictions and limitations imposed by their LLTC had a significant impact on engaging in intimate and sexual relationships. Chronic ill-health may delay sexual or pubertal development and/or growth and accelerate neurodegenerative conditions.

Young adults wished to ensure that sexuality, intimacy, relationships and reproductive considerations were a 'normal' part of growing up. Many young people stated that they felt that they had missed out on the 'experimentation stage' of emerging adulthood, experienced by their non-disabled peers. Most participants were now in their mid-twenties or older, with little or no sexual experiences and limited access to appropriate information and face-to-face peer support.

Question 4: What are the views of partners, parents, carers and care practitioners, who support people with LLTCs, on all of these subjects?

All four groups of participants highlighted the marginalised opportunities of people with LLTCs to experience/demonstrate independent choices about where they live and how and when they socialise with their peers, thus limiting the young adults' opportunities to achieve full adult citizenship and, in particular, sexual intimacy and sexual citizenship (see Chapter 2). While theory and policy statements point the way to new constructions that might increase scope for young people to choose post-school options in dialogue and sexual agency, achieving increased choice calls for social environments to facilitate social participation outside of the health and social care arenas.

Whilst young adults were attending school, college or a hospice, they were part of a peer group. Once they left the education and NHS children's and/or hospice care systems, the young adults spent a lot of time at home, with few face-to-face encounters with their peers, largely relying on social media to keep in touch with both new and former friends.

For health and social care practitioners, support and advice given to young adults about their emerging sexuality is as important as the advice the care professionals receive. This may be optimised and influenced by good and less good practice in the field. GPs and nurse practitioners stated that specialist sexuality training related to palliative care and, more specifically LLTCs, should be part of specialist palliative care training, practice and care plans.

8.3 *The meta-learning*

This research included a number of methodological investigation points which might be referred to as important learning themes (meta-learning findings, see Appendix A), which I have separated from the original research questions and findings discussed in Section 8.1. Methodologically, this thesis has raised two specific considerations meriting attention: posthumous consent, and to changes in regulations and policy have either changed/are changing since I began this research.

Posthumous consent

Informed consent is an essential component of responsible research practice. Whilst the primary researcher may be able to seek consent for their own study, it is impossible to predict how data may be used and applied in future research, particularly if a participant dies during the course of the original research. Two young adults have died since I began this research. During the consent process, one young

man proactively asked me “What will happen to my data if I die during or following your research?” to which I replied, “what would you like to see happen?” The young man responded that he wanted his data to be used after his death to help contribute to knowledge, and he did not set any time limits on its future usage. Between us, we included an additional hand-written sentence to his consent form reflecting his request, which the young man duly signed before his interview. His parents were aware of his participation in my research. The young man had told them after his interview. They were not entirely happy with the decision for his data to be used in this research, and following his death, they contacted me. They respected their son’s request to use his data contribution in this research. I agreed to send the parents a

I did not feel I could ask all participants about posthumous consent (discussed in Chapter 3). Yet, for consistency, I probably should have done. Furthermore, my consent guidance for participants (see Appendices E, F and G) acknowledged that the data would ultimately be ‘deleted after the research is completed and the research findings are published’. This was customary wording at the time of my research ethics applications. I recognise that ‘until the findings are published’ noted in Appendices E, F and G might infer an indefinite period of time, but not necessarily in perpetuity. Most of us do not want to think about death and dying until we have to. Unfortunately, that often means that families are left struggling with difficult decisions about important matters, such as whether the young adult ever made it clear during life what they wanted for themselves after they died. A Living Will or Power of Attorney might be a useful consideration for this potential situation and to its circumstances highlighted by this research.

It is difficult to discuss consent and the use of data following a participant’s death whilst that individual is still living. This is exacerbated if the participants have complex LLCs and the research is of a sensitive nature (see Chapter 3). The literature on posthumous consent primarily relates to reproduction and the use of sperm and egg

donation posthumously (for example, Tremellen and Savulescu, 2015; Jones and Gillett, 2008; Douglass and Daniels, 2002) as well as the complex ethical, moral and legal concerns that have arisen as a result of posthumous assisted reproduction (De Than, 2015). In such cases, guidance has been produced by the Human Fertilisation and Embryology Authority (HFEA).

Researchers owe a duty of care to participants to minimise harm, provide information to seek informed consent, and protect confidentiality. Researchers also have a responsibility within the academic community to maintain professional standards of conduct with transparency and integrity. There is also a duty to produce quality research of wider social value, particularly when and if this involves the use of public funds (Harris, 2018). Many participants may not wish to have their data scrutinised by strangers, particularly following their death. Seeking permission to re-use data following death may reduce the trust between the researcher and potential study participants. This could jeopardise research recruitment for future studies, which may be detrimental to improving the practice evidence base that researchers (Harris, 2018).

Advance directives are important tools for many people, because even the healthiest person could experience a sudden accident and not be able to speak for themselves. However, when a person has a LLTC, it is particularly important to make clear, in writing, one's wishes should a time come when that person cannot express them, and this might include requests about what happens to their research contribution(s). The issues of posthumous consent link closely with the importance of data management.

Data management and repositories

At the end of my doctoral journey, new regulations are coming into force which change how we shall now store and share data. Research data repositories (RDRs) are

storage centres where data can be submitted, stored, checked, scrutinised and subsequently accessed for secondary purposes beyond the original design and design of the initial study. The use of RDRs is now required by academic institutions, publishers and national directives (Harris, 2018). The essence of General Data Protection Regulation (GDPR) is strengthening participants' rights with regard to their data. Acceptable data preservation and sharing has to work with GDPR and this will be built upon in future research. When I circulate my final report to participants, I shall include an update about data storage following discussion with The Data Repository's advisory services at The Open University.

Access to research data increases the impact, efficiency and effectiveness of scientific activities and funding opportunities (Lee and Stvilia, 2017, cited in Harris, 2018). Research repositories provide the opportunity to check and verify research findings, increased transparency of research procedures, and avoid duplication and over-burdening research participants. Young people's palliative care research is an area where the findings may be particularly poignant for research participants (Harris, 2018). The research data may reveal sensitive, contextual information about intimacy, relationships and sexuality, as well as about individuals, family members, and decisions may occur at a difficult transition point for people on an uncertain life course.

Three options have been suggested to protect participants' research data (Bishop, 2009, cited in Harris, 2018). The first is to anonymise data beyond recognition but this may prove difficult, with a potentially small sample of people with LLTCs. The second is to seek consent for the storage and potential re-use of the data if this has not been negotiated or agreed at the outset of the study with participants. The third is to control or restrict access by others to data repositories and carefully manage and monitor the use of data in the future (Bishop, 2014, 2009; cited in Harris, 2018),

possibly limiting access to the original researchers. Again, this may prove difficult as researchers change roles, institutions, retire or will die.

There may be particular challenges for researchers in the arena of young people's palliative care and sexuality research. Anonymisation of data can be difficult in a small but albeit growing number of young adults with LLTCs. People with rare conditions or unusual social circumstances may be identifiable from small amounts of contextual detail. Individual clinical scenarios may not only identify the young adult and family members, but also staff members who provide care and support; and where readership of research outputs is small and interconnected (Harris, 2018). Balancing the aforementioned with removing all contextual information may dilute the value of the original data.

At the time of writing, I am unaware of evidence about the re-use of data without contextual information, particularly in young people's/young adults' palliative care and sexuality research. This topic is extremely challenging and requires careful attention, particularly in relation to the sexuality data I have generated. Whilst completing this thesis, I have sought advice and clarification from the OU Research Ethics Committee and OU Library services on these matters. Both departments agree that the most important consideration for this research is that the conditions of the original information and consent are followed. Preserving and, where appropriate, sharing research data is and will in future be considered 'best practice' and consent forms, as a general principle, will not include 'agreeing *unnecessarily* to destroy data'. The OU Library Services has agreed to provide me with further advice and support where data of a sensitive nature should *be excluded* as well as appropriate preparation of data that might be used for storage and publications.

8.4 Limitations of this study

If I were beginning this research journey again, would my approach differ? Some areas might, because I have learned as I progressed my doctorate and future research would benefit from that learning. I acknowledge that there are limitations to this research, particularly in relation to sampling and diversity (as discussed in Chapter 3). However, I believe that the findings and conclusions hold considerable validity for the following reasons. My research effort adhered to methodological orthodoxy in that I sought to minimise bias among my selection of participants, that I strove for balance and consistency of approach, and that I analysed and interpreted their contributions systematically and transparently. As far as the first of these reasons is concerned, that of bias, there are limitations that may be of interest to the reader.

The views of 35 people; 13 young adults with LLTCs, two partners, ten parents and ten care practitioners were represented. It was my intention to recruit equal, or near equal, numbers of female and male young adults (see Chapter 3). However, of the 13 young adult participants, nine participants were male. It was harder to recruit young adult women, although I wanted to provide gender balance in this research and made considerable efforts to include more women who had mental capacity.

As noted in Chapter three, I was advised by the gatekeepers that many of the young women in the hospices had moderate to severe learning difficulties, requiring assessment under the Mental Capacity Act (2005) and did not meet The Open University's HREC inclusion criteria. I was unable to assess the cognition of potential participants directly in advance of interviews as my access was negotiated through gatekeepers and I was advised by them that participants who contributed were deemed competent to participate. I did not include people with cognitive or learning disabilities because similar research was being undertaken within our faculty and I believed that this study would be rich and comprehensive focussing on people with

LLTCs who had mental capacity (see Chapter 3). Although, the views of people with cognitive impairments were not represented here, there is scope to include them in future research. I recognise that this study only represents the experiences of people *without* cognitive or learning difficulties and that the experiences of those who do have cognitive impairment or learning disabilities are likely to differ.

Despite best efforts, recruiting the fathers of people with LLTCs also proved difficult, most preferring their wives or female partners to engage in this research. Samuel (2014) noted in his research with fathers of disabled children that the experiences of mothers of children with disabilities has been well documented, but that the experiences of fathers remain under-researched. There is often a higher divorce or separation rate in families where there is a child with a disability. Fathers are often excluded from research simply because they are not around or living in the family home (Clarke and McKay, 2008; Beresford and Oldman, 2000). This was the case for two young adults and one parent interviewed in this research (*Lily*, *YAF* and *Liliana PF*). A rounded perspective requires research that includes both parents. Further research in the area would be valuable, to explore the experiences of fathers, including those from ethnic minorities and fathers of young adults with LLTCs, to provide a greater understanding and guidance for supporters and care practitioners (Carpenter, 2008).

All the young adult participants' identified as heterosexual and cisgender therefore limiting the findings of this research to a broadly similar sexual identity and orientation. This was not planned, and every effort was made via the hospices to include people identifying as LGBTQ+. Future research should aim to include individuals who identify as LGBTQ+, see Appendix A, since such individuals are likely to have experiences that differ from the young adult participants represented here. In future research, I would ensure that research briefings and fliers include additional information about the important contribution of people who identify as LGBTQ+.

I had originally sought access to five research sites as I felt this would maximise opportunities for recruitment. Despite best efforts I was only able to recruit from two research sites. This may have impacted on the fact that I recruited less women than originally anticipated and that I was not able to include any fathers.

8.5 *Recommendations for research, policy and practice*

The main findings in this research, discussed in Sections 8.1 and 8.2, indicate a number of recommendations relevant to policy and practice. These have implications for young adults, as well as parents, partners and practitioners, see Figure 4.

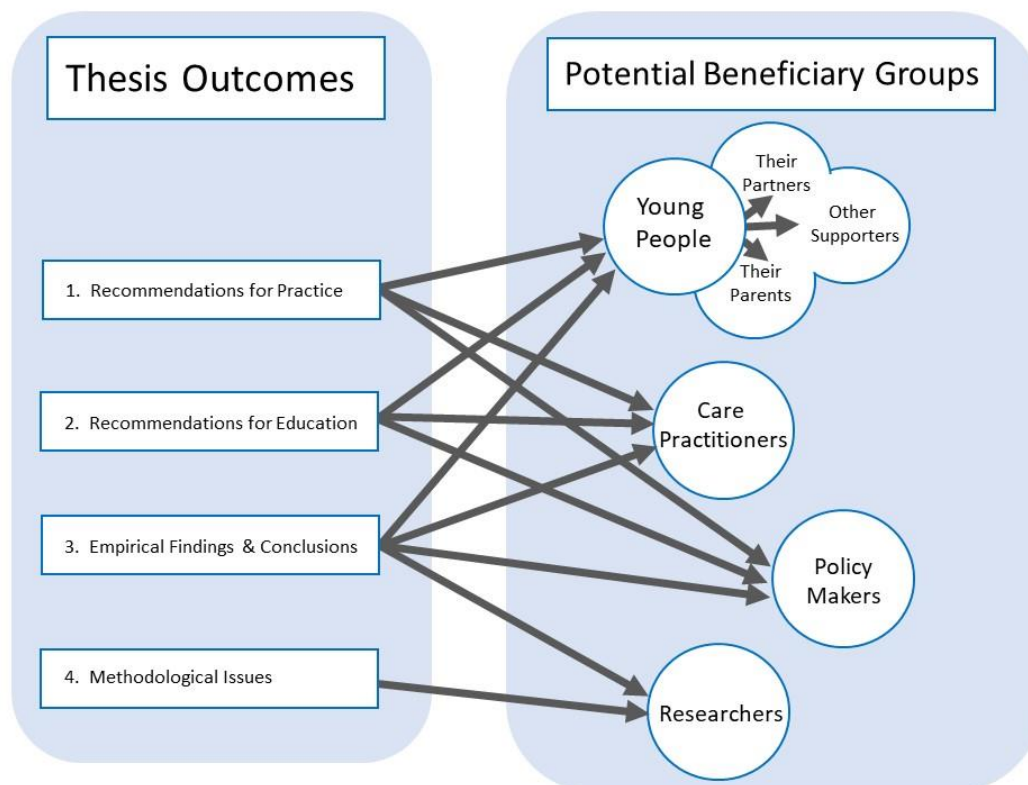


Figure 4: The impact of the study and recommendations for future research, for practice and for education

Importantly, the research has also highlighted some important methodological recommendations.

Recommendation one: Comprehensive information regarding data repositories for research participants

The lack of appropriate information or explanatory material for the public as well as health and social care practitioners about sex(uality) should be addressed as soon as possible. Further research to enable a fully informed debate about consent and research data repositories (RDRs) (which came into force at the end of May 2018) and should be ongoing as part of RDR development.

The following approaches might be applied to sensitive data management, such as my own: First, to restrict access to the data repositories to the researchers of the original study, within the boundaries of the initial consent or assent agreements. Second, limiting the data that may be accessible, such as meta-data. Third, by limiting access 'by time', such as having an embargo in place for a specified period but not in perpetuity. These arrangements will need enforceable license agreements in place to specify how the data will be used, that it will not be disseminated further, and that confidentiality will be respected. Information on how decisions are made, enforced, or who is accountable for the appropriate use of RDR is required. Furthermore, information to explain RDR to lay members of the public as part of the consent process is required.

Recommendation two: Posthumous consent

Due consideration should be given as to how researchers seek consent from research participants to use data following their death. This should not be limited to drawing upon data related to sperm donorship but include other groups where the data

generated during research may be of importance, such as young people's palliative care and the sexuality and disability research.

Recommendation three: Gatekeeping

To avoid delay and disappointment, early contact and dialogue between the researcher and potential gatekeepers is imperative.

Empirical findings: Implications for policy, practice and education

Recommendation four: Transition services for young adults with LLTCs

Sustainable, dedicated, transition services for young adults with LLTCs are required that address their complex and holistic requirements, including services that address the young adult's sex(uality), such as that designed by Morgan Blackburn and Bax, (1995). It is important that the social, sexual and emotional needs of young people with LLTCs are fully met. Young adults with LLTCs, their families and carers, must be recognised as a discrete group and be appropriately supported with their health and social needs, including sex(uality).

Recommendation five: Specialist sex education for young adults with LLTCs

Access to specialist information and support about sex(uality) and the specific issues related to individual LLTCs (such as duchenne muscular dystrophy and cystic fibrosis) should be available for young adults with LLTCs and their supporters (parents), as required), during school, college and throughout life.

Recommendation six: The development of online education resources (OERs) related to the sexuality of young adults with LLTCs, in order to increase public awareness, such as those produced by The Open University.

Recommendation seven: Specialist sex education for nurses, doctors and other healthcare practitioners

There is a need for specialist sex education to ensure that health and social care practitioners are confident and competent in addressing the sexuality of people with LLTCs. Specific specialist training should be available at both undergraduate and postgraduate levels for nurses, doctors and other health and social care practitioners.

Recommendation eight: Genetic screening and counselling

Access to genetic screening and counselling should be available for young adults with LLTCs, their partners and their parents.

Recommendation nine: Guidance and governance

Clear guidance and governance in hospices should be available as to how young adults can discuss and manage sexual choices in residential settings.

Recommendation ten:

Further research with young adults with LLTC who are parents or who have experience(d) pregnancy or wish to have a child.

Recommendation eleven: Further research into the sexual experiences of young adults with LLTCs.

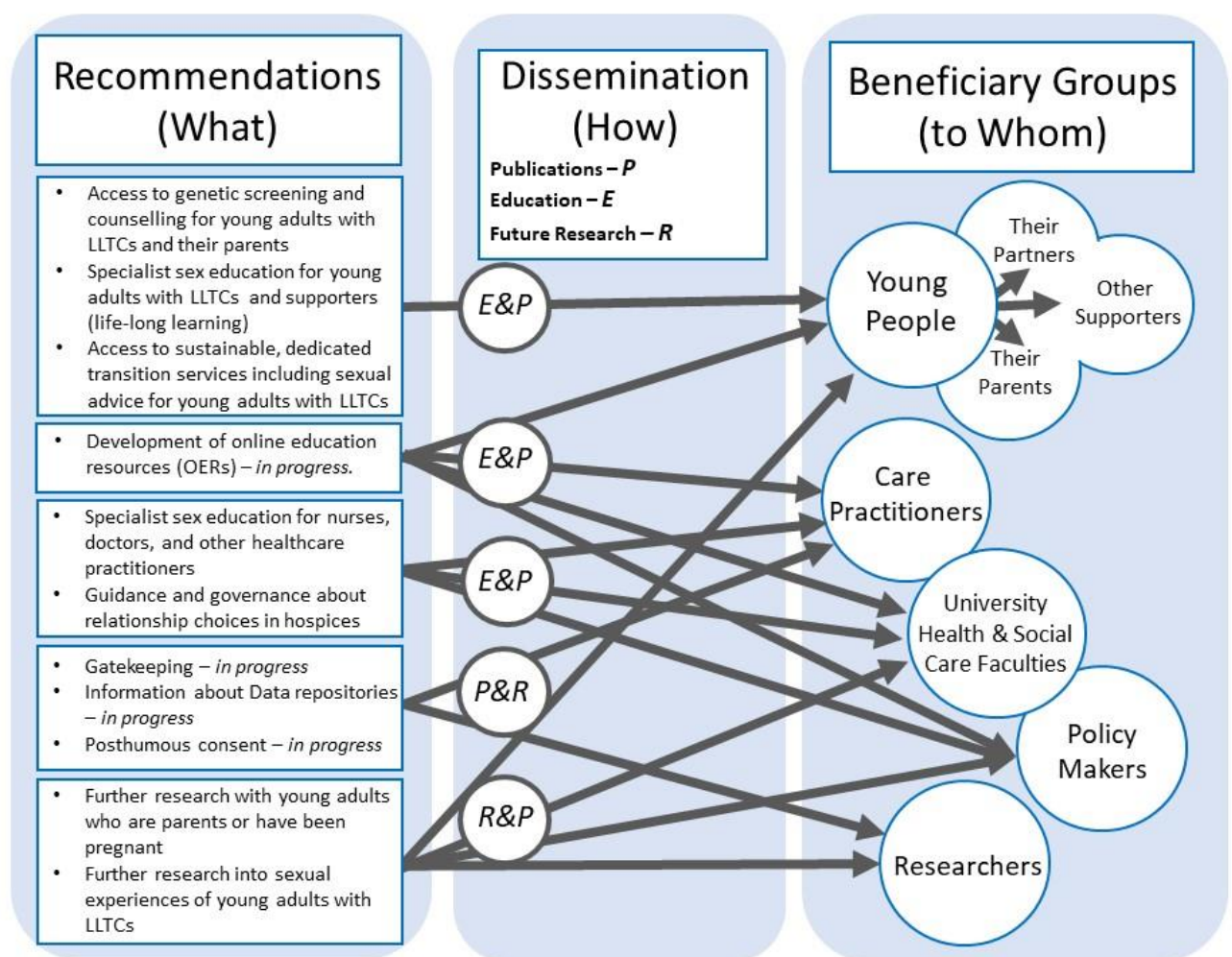


Figure 5: A dissemination plan, outlining intentions including publications, future research and suggestions for education.

8.6 *Final reflections*

Reflections on my doctoral journey

A central feature of this thesis was the privilege of engaging in dialogue with participants about a previously neglected subject, but which is a developing and important area of research (Liddiard, 2018). The interviews and analysis looked at intimate stories (Plummer, 2000), of the sex(uality) of young adults with LLTCs and the particular issues for partners, families and care practitioners supporting them. This close examination of the research encounter encouraged me to question some of the emerging methodological wisdoms about research with young adults with disabilities and the role of the researcher.

This thesis has explored the opportunities for, and experiences of, relationships, intimacy and sex, as well as the reproductive aspirations and choices in young adults aged 16 years and over with life-limiting and life-threatening conditions, in England, using a life course perspective and drawing on the concepts of uncertainty, liminality, transition points and biographical disruption. This perspective has provided a degree of flexibility in a highly complex field, whilst logically remaining connected to the research but not changing its emphasis or focus (Stenner et al, 2017). The life course perspective is increasingly recognised as an important multidisciplinary framework for the study of health, illness and mortality (Denny, Earle and Hewison, 2016).

I did not want to take things at face value; rather I wanted to explore in depth the opportunities and experiences of sex(uality) of young adults with LLTCs and what the presence and absence of any gaps meant to these young adults and their supporters. I discussed the need for a reflexive approach to this type of qualitative study in

Chapters 1 and 3. Here my reflection, focuses on the thesis experience, the ambition, the research questions, the findings and some of the challenges in undertaking a thesis later in life.

One of the positives of writing a thesis was the significant benefit that accrued from being able to reflect from a perspective of spending a significant part of my professional life working in the specialist area of young people's palliative care.

With the benefit of hindsight, I realise that this research task was large. Interviewing four different groups of participants face-to-face, and some people more than once, around England, was perhaps naively ambitious within the timescale. No doubt this is not an unusual comment in the concluding stages of a doctorate (Barbour, 2014).

The research questions were open-ended, thus requiring divergent exploration rather than convergent decision-making with all groups of participants. In all four interview questions, I drew upon the sex(uality) shorthand described in Chapter 1, to explore meanings, information, experiences and reproductive choices with the participants. This provided an *aide memoire* and helped me to keep track of the questions I was asking. However, this was not obvious to the participants. The young adults with LLTCs were the primary focus of this research; I was seeking their views, understandings, attitudes, aspirations, opportunities and experiences of sex(uality) and reproduction. Importantly, the contributions of their parents and care practitioners often validated their views, but also inevitably provided differing perspectives (see Chapters 4 to 7).

I have attempted to accurately recount the meanings and stories from all four groups of participants, in their own words. None of the questions were likely to evoke simple answers, but rather to highlight the complexity of the young person's LLTC(s), balancing this with their wishes and feelings about relationships and intimacy, whatever that meant to them: to be loved and to give love, to be intimate, to be

cuddled, touched, smelt or just 'to have sex' (*Jane, YFP*) and, for one couple, possibly to parent a child (*Vincent, YMP*, and *Smidge, FP*) (see Section 7.2). The variety of the participants narratives I encountered was one of the many rich tapestries of this research and I am grateful to all the participants.

In writing this thesis I had to make choices about which data to include and to exclude, which was a challenge when all the data seemed important. Careful data selection was imperative in order not to dilute the importance of the key empirical findings in this research. It is worth reflecting on what I excluded and why. Much of the data included framed the most significant findings, discussed in Chapters 4 to 7, and were closely linked to the participants' profiles, outlined in Chapter 3 and Appendix I.

The process of fieldwork, analysis and writing up has involved a range of research activities and this generated feelings. One of the questions that underpinned my exploration, and part of the rationale of exploring sex(uality), was why this is such a taboo and sensitive subject. I recognised that doing 'sensitive' fieldwork would evoke memories of people who were unwell, dying, had lost siblings or children, or were unable to have children; all deeply significant losses. A particularly difficult aspect for me as a clinician was the intensity of the professional research relationship during the fieldwork phases and the challenges of concluding the research relationship with participants at the end of the study.

I was conscious of what I brought to the research relationship, and needed to separate this, as well as myself, from the findings and this was not easy. Mindful of this, I arranged clinical supervision sessions based on those I had experienced in my work in the NHS and the third sector, but incorporating a model described by Hunt, Swallow and Twycross (2014). Their model is used for doctoral candidates, aimed at providing peer support for nurses who are researching 'sensitive' areas related to children and young people and who wish to discuss the specific ethical challenges

encountered during fieldwork and throughout the doctoral journey. Similarly, towards the end of my own fieldwork, The Open University set up a faculty 'PhD Fieldwork Facilitation Group', led by a researcher not supervising attendees. The aim of this group was to discuss ethical challenges encountered during fieldwork, under Chatham House Rules, see Appendix A. Prior to this group being established, with the approval of my academic supervisors, I sought the assistance of an independent mentor who complemented but did not conflate or contradict my supervisors' academic advice. With her, I discussed the impact of the fieldwork, the sensitivity of addressing sex(uality), death and dying, , focusing on the ethical issues that had arisen during fieldwork and data analysis. Being able to talk about some of the ethical dilemmas and challenges of this research in a neutral space and in confidence helped me to focus on the sociological aspects of what was taking place and to appropriately utilise my academic supervision sessions to focus on the key research activities and deliverables.

At times, like many researchers, I found the analytical process both poignant and stressful. Listening to the audio tapes, personally transcribing the interviews and subsequently reading the transcripts many times, importing them into the qualitative software, NVIVO 10, and Excel spreadsheets, and then analysing the data was all-absorbing and time consuming. Listening to the voice recordings of two (now) deceased individuals was particularly moving. In addition, I was concerned that the young people and parents in this research were constantly explaining their LLTCs to others as well as to me in this research. I was also asking participants about sex(uality), often regarded as a taboo subject, to assist in coding my data and to reach some conclusions based on the interviews with each participant.

The experience of my fieldwork remained long after completing the analysis. This is inevitable as one confronts the interview data at various stages of the thesis 'write up', which does not end at the point of interview. It might seem obvious that I was drawn to this research because of my previous policy work in the palliative care sector.

Researchers who work in a sensitive area are expected to be able to cope with sensitive topics such as sex(uality), uncertainty around the life course, dying and death, and this is important to try and make academic sense of what is taking place.

I began this thesis journey with an explanation of why I wanted to explore relationships, intimacy and the sex(uality) and reproduction of young people aged 16 years and over with LLTCs. Essentially, I believed that it was timely to explore the sex(uality) of an emerging and growing population of people with LLTCs who, until recently, were not expected to live into adulthood.

As stated, in Chapter 3, over a two-year period, I collected data via face-to-face interviews with 35 participants, including adults with LLTCs, and their parents, partners and care practitioners. To validate the study and thus provide the rich accounts of sex(uality) across an uncertain life course, the data which I have presented in this thesis has not only highlighted the gap between rhetoric, policy and practice, but presented some of the stigmas and taboos of sex(uality) at certain transition points within an uncertain life course. However, a key finding in this research is that young adults with LLTCs are marginalised and denied a fundamental right of sexual citizenship, discussed in Chapters 5 and 6. Throughout this thesis I have drawn upon sex(uality) literature to help me to interpret the data and the life course perspective, which supported the thematic analysis of my data.

Reflections on the findings

Given their roles and life-long caring responsibilities, I might have expected more parents to appear overprotective of their sons or daughters, and care practitioners to express different views from the young person's and to 'shy away' from discussing sex(uality) in what is often viewed as a 'taboo subject'. In reality, this was not always the case. *Topaz*, *Liliana* and *Goldie (MF)* were particularly supportive that *Lily* and

Vincent (YAs) and Goldie's son should receive the necessary support and encouragement to engage in relationships and be intimate

There was a degree of reticence and insecurity among care practitioners about discussing sexual facilitation and as to where and whether sexual facilitation was sanctioned in hospices, highlighting moral values guiding service provision. I also question whether my being an older woman may have inhibited discussion by younger participants about sex(uality). My previous sexuality research had taught me how to engage in discussions about sexuality with disabled people. I am not easily embarrassed and am receptive to the legal and ethical sensitivities of some discussions.

A period of ill health and family bereavements required me to suspend my doctoral research for over a year. The major downside to this being, when I returned to study, I felt I should re-analyse and re-check all the data. This was not an easy task, checking and re-checking over 1000 pages of manuscript on NVIVO, and re-checking digital recordings with hundreds of codes. However, the gap in study provided renewed rigour and further reflection about the data. Whilst the coding and re-analysis did not change significantly, further themes emerged, particularly related to the uncertainty around the pivotal moments of diagnosis, discussed in Chapter 4.

Reflections about the co-researcher model

I did not use a co-researcher model in this research. The reflective debate here is if I began my research now, would I adopt this methodological model again, or with the benefit of learning and hindsight, use a co-researcher model? Inclusive or co-research is developing and is becoming a research model of choice with disabled people. Ellis (2015) engaged seven adults with learning disabilities as co-researchers in her doctoral

research. Notwithstanding, Ellis (2015) discusses the many challenges to ‘co-researching’, and that some of these may minimise a fully inclusive research approach.

There were several reasons why I did not use the co-researcher model. First, and importantly, was a degree of time sensitivity which required me to adopt a focused approach and to maintain and manage my research effort efficiently, particularly as this included a larger cohort of participants than in Ellis’s (2015) research. She also acknowledges that the timing and preparation is considerable and compromises in the decision-making process may have to be considered (Ellis, 2017).

My second consideration related to the potential demands on the research participants with complex and debilitating LLTCs. I questioned the ethics of engaging young adults who might only have a short time left to live, and coping with the additional demands of a co-research agenda may prove an added burden. The research questions made demands on participants. In my research I was already asking a lot of individuals by inviting them to share their views about a sensitive topic, sexuality. Ellis (2015) also noted that researchers sometimes assume that participants will want to be co-researchers and be involved in research, but this is not always the case. Some co-researchers may prefer to adopt a more distanced role, such as commenting online about research questions and design.

My third reason specifically related to resources and risk assessment. In a separate project, Blackburn, Chambers and Earle (2016), young adults with LLTCs were consulted throughout that study about the design, content and development of sexuality resources for health and social care practitioners (The Open University, 2016). By definition, they were not co-researchers. Their engagement was through focus group discussions, usually held in fully accessible hospice sites where care staff were readily available if assistance was required or someone became unwell. Enabling young adults to be co-researchers is resource intensive, not only of time, but in terms

of the important advanced planning required if people are away from their usual place of residence. People with LLTCs, for example, may require ventilation, suction, parenteral nutrition (TPN), see Appendix A, or continence management. Thus, a deterrent was the significant risk assessment requirements before scheduling meetings at venues where back-up medical facilities and equipment were unavailable.

Consequently, my research effort was designed, predicated and determined by the discipline of finite resources. On the doctoral journey, part of my learning was the art of managing the research. The co-researcher model required a level of expertise that I did not think I had or could manage as a solo-researcher. My decision was to keep the focus and momentum of this research realistic, not overly ambitious and to minimise the additional complexity of managing a collaborative group of co-researchers.

Moreover, my focus was on an area of sexuality research: people with LLTCs. Had I opened this up to co-research, the focus might have shifted. Much innovative, inclusive research is very methodologically orientated, so it becomes more about the process rather than the outcomes (Ellis, 2017). I wanted this research to focus on the outcomes and contribution to knowledge about a very specific, niche area, and not necessarily on the methodological innovations of co-research.

Notwithstanding, I fully acknowledge the importance of inclusive research, and feel to some extent that some participation has been achieved through regular dialogue and communication with participants. To conclude this debate, should further research be adopted with a similar group of participants, I would consider pursuing this route earlier, aiming to make possible what was previously impossible for potential co-researchers and myself, having first considered the points, a priori, I have outlined in this section.

8.7 Conclusion

This thesis has provided a rare opportunity to understand and ask critical questions about the widely held assumptions about the sex(uality) of an emerging and growing population of young adults with LLTCs. As stated throughout, young adults with LLTCs were not previously expected to reach adulthood. I have argued that the exploration of this group of people's sex(uality) is a new area of research. As such, it provides a hitherto, rarely addressed, cross-section exploration of the sexuality of this population whose overall life-expectancy is increasing. This opens the door to further research on the sex(uality) of individual groups of people with LLTCs, particularly those with rare LLTCs.

I have shared some of the complications and difficulties encountered in terminology and the organisation of hospice and specialist palliative care, identifying a liminal space and gap between children's and adult palliative care provision: the holistic requirements of young adults with LLTCs. I have substantiated my assertion that young adults with LLTCs are a new and emerging population. In terms of hospice provision, children's versus adults' palliative care provision, and young people aged over 16 years, there emerges an ambiguous 'liminal group', somewhere in between adolescence and older adults, whose needs can no longer be ignored and who require specialist recognition and services.

Sexual expression and intimacy is one of the most fundamental of human pleasures and is important, regardless of whether or not a person has a LLTC. Sexual intimacy, in whatever form, is an integral part of a person's emotional and physical well-being. The sexual citizenship of people with LLTCs is not just a human right; it is an issue of social justice that must not be ignored.

References

Abbott, D. W. F. (2012) 'Other voices, other rooms: Talking to young men with Duchenne muscular dystrophy (DMD) about the transition to adulthood, *Children and Society*, vol. 26, no. 3, pp. 241–50.

Abbott, D. W. F. (2015) 'Love in a cold climate: changes in the fortunes of LGBT men and women with learning disabilities?' *British Journal of Learning Disabilities*, vol. 43, pp. 100–105.

Abbott, D. W. F. and Carpenter, J. S. W. (2012) 'Talking about tricky transitions with young men with Duchenne muscular dystrophy and their families', *BMJ Supportive and Palliative Care*, vol. 2, no. 2, pp. 178–179.

Abbott, D. W. F. and Carpenter, J. S. W. (2014) "'Wasting precious time": Young men with Duchenne muscular dystrophy negotiate the transition to adulthood', *Disability and Society*, vol. 29, pp. 1192–1205.

Abbott, D. W. F. and Carpenter, J. S. W. (2016) "The things that are inside of you are horrible": Children and young men with Duchenne muscular dystrophy talk about the impact of living with a long-term condition', in Kelly, B. and Byrne, B. (eds) *Valuing Disabled Children and Young People: Research, Policy, and Practice*, Routledge, pp. 143–153.

Abbott, D. W. F., Carpenter, J. S. W. and Bushby, C. (2012) 'Transition to adulthood for young men with Duchenne muscular dystrophy: Research from the UK', *Neuromuscular Disorders*, vol. 22, no. 5, pp. 445–456.

Abbott, D. W. F., Jepson, M. and Hastie, J. (2016) 'Men living with long term conditions: Exploring gender and improving social care', *Health and Social Care in the Community*, vol. 24, pp. 420–427.

Abbott, D. W. F., Prescott, H., Forbes, K., Fraser, J. and Majumdar, A. (2017) 'Men with Duchenne muscular dystrophy and end of life planning', *Neuromuscular Disorders*, vol. 27, pp. 38–44.

ACT and Triangle (2011) 'The whole picture: Exploring disability and sexuality' [Video]. www.togetherforshortlives.org.uk/professionals/resources/2444_lets_talk_about_sex (Accessed 6 October 2015).

Aldridge, J. (2008) 'Living with a Seriously Ill Child', *Sheldon Press*.

Aldridge, J., Shimmon, K., Miller, M., Fraser, L. K. and Wright, B. J. D. (2017) "'I can't tell my child they are dying': Helping parents have conversations with their child", *Archives of Disease in Childhood*.

Amery, J. M. (2016). 'A practical guide to children's and young people's palliative care', Lulu Publishing Services (Accessed on 19/02/2016)

Anderson, E.M. and Clark, L. (1982) *Disability in Adolescence*. London Methuen.

Arnett, J. J. (2000) 'Emerging adulthood: A theory of development from the late teens through the twenties', *American Psychologist*, vol. 55, no. 5, p. 477.

Aronson, J. (1994). 'A Pragmatic View of Thematic Analysis. The Qualitative Report', 2(1), 1-3. Retrieved from <https://nsuworks.nova.edu/tqr/vol2/iss1/3> (Accessed March 10th 2018).

Balmer, C., Griffiths, F. and Dunn, J. (2015) A 'new normal': exploring the disruption of a poor prognostic cancer diagnosis using interviews and participants-produced photographs, *Health Vol.* 19, No. 5, pp.451–72.

Bamforth, N. (2012) 'Sexuality and citizenship in contemporary constitutional argument', *International Journal of Constitutional Law*, vol. 10, no. 2, pp. 477–492, Oxford University Press. On-line <https://doi.org/10.1093/icon/mor065> (Accessed 20th March 2018)

Barbour, R. (2010) 'Using qualitative methods in comparative research', *Salute e Società*, vol. 9, no. 2, pp. 65–79.

Barbour, R. (2013) 'Doing Focus Groups.' *The Sage Qualitative Research Kit*. London.

Barbour, R. (2014) 'Introducing Qualitative Research', London, Sage.

Barton, L. (2005) 'Emancipatory research and disabled people: some observations and questions', *Educational Review*, vol. 57, no. 3, pp. 317–327.

Bauer, M.W and Gaskell, G. (2000) 'Qualitative researching with text, image and sound', *A practical handbook*. Sage. London

Beecham, J., Snell, T., Perkins, M., Knapp, M. (2008) 'After transition: Health and social care needs of young adults with long-term neurological conditions;. PSSRU Research Summary 48, Bristol, UK: PSSRU *Policy and Practice*, Doctoral thesis, UCL.

Beale, E. A., Baile, W. F. and Aaron, J. (2005) 'Silence is not golden: Communicating with children dying from cancer', *Journal of Clinical Oncology*, vol. 23, no. 15, pp. 3629–31.

Bell, D. (1997) 'Sex lives and audiotape: Geography, sexuality and undergraduate dissertations', *Arena Symposium: Teaching Sexual Geographies*, vol. 21, no. 3, pp. 411–417.

Bell, D. and Binnie, J. (2000) 'The Sexual Citizen: Queer Politics and Beyond', *Cambridge: Polity Press*.

Bennet de Marrais, K. (1998) 'Inside Stories: Qualitative Research Reflections,' *London, Routledge*.

Beresford, B. A. and Stuttard, L. (2014) 'Young adults as users of adult healthcare: experiences of young adults with complex or life-limiting conditions', *Clinical Medicine*, vol. 14, no. 4, pp. 404–408 [Online]. DOI: [10.7861/clinmedicine.14-4-404](https://doi.org/10.7861/clinmedicine.14-4-404) (Accessed 16 December 2018).

Beresford, B. (2004) 'On the road to nowhere? Young disabled people and transition', *Child: Care, Health and Development*, vol. 30, no. 6, pp. 581–587 [Online]. DOI: [10.1111/j.1365-2214.2004.00469.x](https://doi.org/10.1111/j.1365-2214.2004.00469.x) (Accessed 2 April 2018).

Beresford, P., Harrison, C. and Wilson, A. (2002) *Mental health service users and disability: implications for future strategies*, Policy and Politics, Vol. 30, pp. 387–96.

Beresford, B. and Oldman, C. (2000). 'Making Homes Fit for Children: Working Together to Promote Change in the Lives of Disabled Children', Bristol: The Policy Press.

Berg, B.L. and Lune, H. (2004) *Qualitative Research Methods for the Social Science*, Pearson.

Bishop, L. (2009) 'Ethical sharing and reuse of qualitative data', *Australian Journal of Social Issues*, Australian Social Policy Association, vol. 44, no. 3, p. 255.

Bishop, L. (2014) 'Re-using qualitative data: A little evidence, on-going issues and modest reflections', *Studia Socjologiczne*, vol. 3, no. 214, pp. 167–176.

Boeijea H., Duijnsteeb M., Grypdonckb M. & Pool A. (2002) 'Encountering the downward phase: biographical work in people with multiple sclerosis living at home'. *Social Science and Medicine* 55(6), 881–893.

Black, B.P., Holditch-Davis, D. and Miles, M.S. (2009) 'Lifecourse theory as a framework to examining becoming a mother of a medically fragile pre-term infant', *Res. Nurse Health*, Vol 32. No.1, pp. 38–9.

Blackburn, M. C. (2002) 'Sexuality and Disability', *Butterworth/Heinemann: Elsevier Science*, ISBN 0-7506-2252-0

Blackburn, M. C. (2007) 'Safeguarding children and young people with continence difficulties', in Bonner, L. and Wells, M. (eds) *Effective Management of Bladder and Bowel Problems in Children*, pp. 5–24, Class Publishing, London

Blackburn, M. C. (2010) '*Children's Hospice Care: A Report Based on Visits to All Children's Hospice Services in the UK; Children's Hospices UK*, Bristol.

Blackburn, M., Chambers, L., Earle, S., De Than, C. and Raeburn, D. (2016) *Talking About Sex, Relationships and Intimacy: Guidance and Standards for Those Working with Young People with Life-Limiting and Life-Threatening Conditions*, The Open University and Together for Short Lives.

Blackburn, M., Chambers, L. and Earle, S. (2016) 'Talking about sex, relationships and intimacy: New guidance and standards for nurses and other health and social care professionals working with young people with life-limiting and life-threatening conditions', *Journal of Advanced Nursing*, vol. 73, no. 10, pp. 2265–2267.

Blackburn, M. C., Earle, S. and Komaromy, C. (2014) 'Relationships and sexuality in young adults with life-limiting conditions in the UK', *BMJ Supportive and Palliative Care*, vol. 4, A27 [Online]. DOI: 10.1136/bmjspcare-2014-000654.74 (Accessed 17 May 2017).

Blakemore SJ, Robbins T. 2012. 'Decision-making in the adolescent brain'. *Nature Neuroscience*. 15 (9): 1184 – 1192. doi:10.1038/nn.3177

Blows, E, Bird, L, Seymour, J and Cox, K (2012) 'Liminality as a framework for understanding the experience of cancer survivorship: a literature review'. *Journal of Advanced Nursing* 68, 2155–2164.

Bogaert, A.F. (2015) 'Asexuality: what it is and why it matters', *Journal of Human Sexuality*, Vol. 52 No. 4, pp. 362–79.

Bourne, A. H. and Robson, M. A. (2013) 'Participants' reflections on being interviewed about sexual behaviour: Implications for collection of qualitative data on sensitive subjects', *International Journal of Social Research Methodology*, vol. 18, no. 1, pp. 105–116.

Braun, V. and Clarke, V. (2006) Using thematic analysis in psychology. *Research in Psychology* 3(2), 77-101.

British Sociological Association (2002) Statement of Ethical Practice. Available at: <https://www.britsoc.co.uk/media/23902/statementofethicalpractice.pdf> (accessed on 29 November 2016).

Brook (2014) *Sex and Relationships Education (SRE) for the 21st Century*, London [Online]. Available at www.brook.org.uk/your-life/category/relationships (Accessed 6 October 2015).

Brown, E., Coad, J., and Franklin, A. (2017) 'The impact of a sibling's life-limiting genetic condition on adult brothers and sisters', *American Journal of Medical Genetics Part A*, vol. 173, no. 7 [Online]. DOI: [10.1002/ajmg.a.38213](https://doi.org/10.1002/ajmg.a.38213) (Accessed 23 March 2018).

Brown, M. and Sourkes, B. (2006) 'Teens who are coping with a life-threatening illness', *ChiPPS Pediatric Palliative Care Newsletter*, no. 5, September.

Brown, P. (1995) 'Naming and framing: The social construction of diagnosis and illness', *Journal of Health and Social Behavior*, extra issue, pp. 34–52.

Browne, K. (2005) 'Snowball sampling: Using social networks to research non-heterosexual women', *International Journal of Social Research Methodology*, vol. 8, no. 1, February, pp. 47–60 [Online]. DOI: [10.1080/1364557032000081663](https://doi.org/10.1080/1364557032000081663) (Accessed 20 January 2018).

Bruner, J. (1990) 'Acts of Meaning', Cambridge MA: *Harvard University Press*.

Bryman, A. (2008) 'Social Research Methods', 3rd edn, Oxford, *Oxford University Press*.

Burne, S. R., Dominica, F. and Baum, J. D. (1984) 'Helen House – a hospice for children: analysis of the first year', *British Medical Journal (Clinical research ed.)*, vol. 289, pp. 1665–1668

Bury, M. (1982) 'Chronic illness as a biographical disruption', *Sociology of Health and Illness*, vol. 4, no. 2, pp. 167–82.

Bury, M. (1991) 'The sociology of chronic illness: a review of research and prospects', *Sociology of Health and Illness*, vol. 13, no. 4, pp. 451–468.

Bury, M. (1997) 'Health and Illness in a Changing Society', *London, Routledge*.

Cairns R. B. and Rodkin, P. C. (1998) 'From configurations to pathways', in Cairns R. B. Bergman, L. R. and Kagan J. (Eds) *Methods and models for studying the individual*, Thousand Oaks CA, Sage, pp. 245–65.

Canaday, M. (2011) *The Straight State: Sexuality and Citizenship in Twentieth-Century America*, Princeton, NJ: Princeton University Press.

Cancer Research UK (2018) <https://www.cancerresearchuk.org/> (Accessed 18 March 2018).

Care Quality Commission (2014) *From the Pond into the Sea: Children's Transition to Adult Health Services* [Online]. Available at: www.cqc.org.uk/sites/default/files/CQC_Transition%20Report.pdf (Accessed 6 October 2015).

Carpenter, B. and C. Towers (2008), *Recognising Fathers: The Needs of Fathers of Children with Disabilities*, *British Journal of Learning Support*, 23 (3), 118-25.

Charmaz, K. (1999) 'Stories of suffering: Subjective tales and research narratives', *Qualitative Health Research*, vol. 9, no. 3, pp. 362–382.

Charmaz, K. (2004) 'Premises, principles, and practices in qualitative research: Revisiting the foundations', *Qualitative Health Research*, vol. 14, no. 7, pp. 976–93.

Charmaz, K. (2017) '*The power of constructivist grounded theory for critical enquiry*', *Qualitative Enquiry*, Vol 23(1), 34–35.

Charmaz, K. and Milligan, M. J. (2006) 'Grief', in Stets, J. E. and Turner, J. H. (eds) *The Handbook of the Sociology of Emotions*, pp. 516–538, New York, Springer.

Clark, D. (1998a) 'Originating a movement: Cicely Saunders and the development of St Christopher's Hospice, 1957–67', *Mortality*, vol. 3, no. 1, pp. 43–63.

Clark, D. (1998b) 'An annotated bibliography of the publications of Cicely Saunders – 1: 1958–67', *Palliative Medicine*, vol. 12, no. 3, pp. 181–193.

Clark, D. (1998c) 'History and Culture in the rise of palliative care'. *Palliative Care* eds. Payne, S., Seymour, J and Ingleton, C, in *Nursing: Principles and evidence for practice*: P.39-44.

Clarke, H. and McKay, S (2008) Exploring disability, family: formation and break-up: Reviewing the evidence: Department for Work and Pensions Research Report No 514.

Clough, P and Nutbrown, C. (2012) '*A Student's Guide to Methodology; Justifying Enquiry*'. Sage publications, ISBN 978-1-4462-0862-5.

Connor, S. R., Downing, J. and Marston, J. (2017) 'Estimating the global need for palliative care for children: A cross-sectional analysis', *Journal of Pain and Symptom Management*, vol. 53, no. 2, pp. 171–177.

Corbin, J.M. and Strauss A. (1991) 'A nursing model for chronic illness management based upon the trajectory framework', *Scholarly Enquiry for Nursing Practice*, Vol. 5 No. 3, pp. 155–74.

Corlyon, J. and McGuire, C. (1997) 'Young Parents in Public Care,' *London, National Children's Bureau*.

Craig, F. and Lidstone, V. (2012) 'Adolescents and young adults', in Goldman, A., Hain, R. and Liben, S. (eds) *Oxford Textbook of Palliative Care for Children*, pp. 90–99.

Crenshaw, K (1989). 'Demarginalizing the intersection of race and sex: a Black feminist critique of antidiscrimination doctrine, feminist theory and antiracist politics'. *University of Chicago Legal Forum*. 1989: 139–168. (Accessed February 10, 2019)

Crotty, M. (1998) 'The Foundations of Social Research'. London *Sage Publications*.

Crouch, N. and McKenzie, H. (2000) 'Social realities of loss and suffering following mastectomy', *Health*, Vol. 4 No. 2 pp. 196–215.

Cystic Fibrosis Trust (2018) Available at www.cysticfibrosis.org.uk (Accessed 20 April 2018).

Davies, D. (2000) 'Sharing our stories, empowering our lives: Don't dis me!', *Sexuality and Disability*, vol. 18, no. 3, pp. 179–18.

Day, E. (2016) 'Principles and Practices for Involving Teenagers in Decision-Making about their Care and Treatment: A Prospective Participant-observation Study to Inform Policy and Practice', *Doctoral thesis, UCL*.

Dean, L., Tolhurst, R., Khanna, R. and Jehan, K. (2017) "'You're disabled, why did you have sex in the first place?' An intersectional analysis of experiences of disabled women with regard to their sexual and reproductive health and rights in Gujarat State, India', *Global Health Action*, vol. 10, no. 2 [Online]. DOI: 10.1080/16549716.2017.1290316 (Accessed 30 March 2018).

Dennison, P.H. (2017) 'The Professionalisation of Teaching in Higher Education in the UK; Perceptions and Understandings, *Thesis, University of Greenwich*.

Denny, E. (2016) 'Long term illness and conditions', in Denny, E., Earle, S., and Hewison, A. (Eds) *Sociology for Nurses*, pp. 160–81.

Denny, E. and Earle, S. (eds) (2009) 'The Sociology of Long term Conditions and Nursing Practice', Houndmills, Basingstoke, Hampshire: *Palgrave Macmillan*.

Denny, E., Earle, S. and Hewison, A (eds) (2016) 'Sociology for Nurses', third edition *Polity Press*

Denzin, N. and Lincoln, Y. (1994) 'Handbook of Qualitative Research', Thousand Oaks (California), *Sage Publications*.

Department for Education (2019a) 'Relationships Education, Relationships and Sex Education, and Health Education in England' *Government consultation response: February 2019*

Department for Education (2019b) 'Introduction of statutory Relationships Education, Relationships and Sex Education and Health Education', *Equality Impact Assessment and Public Sector Equality Duty*. February 2019 updated.

De Than, C. (2014) 'Sex, disability and human rights: Current legal and practical problems', in Owen, T. and De Than, C. (eds) *Supporting Disabled People with their Sexual Lives: A Clear Guide for Health and Social Care Professionals*, Jessica Kingsley Publishers.

De Than, C. (2015) *Sex, Disability and Human Rights: Current Legal and Practical Problems*, London, Jessica Kingsley Publishers, pp. 86–102.

De Than, C. and Elvin, J. D. (2016) 'The relationship between capacity and consent', in Reed, A. and Bohlander, M. (eds) *Consent: Domestic and Comparative Perspectives*, pp. 37–51, Routledge.

Devanney, C. and Bradley, S. (2012) 'Count Me In: Children's Hospice Service Provision 2011/12', Durham University Mapping Team with Together for Short Lives.

Dickson-Swift, V., James, E. L. and Liamputtong, P. (2008) 'Undertaking Sensitive Research in the Health and Social Sciences: Managing Boundaries, Emotions and Risks', *Cambridge University Press*, p. 168.

Diener, E. and Crandall, R. (1978) 'Ethics in Social and Behavioural Research', *University of Chicago Press*, Chicago.

Dilorio, C., Kelley, M. and Hockenbury-Eton, M. (2013) 'Communication about sexual issues: mothers, fathers and friends', *Journal of Adolescent Health*, Elsevier, Vol 24, No. 3, pp. 181–89.

The Disclosure and Barring Service (DBS) (2013). Available at <https://www.crb-check.com/> (Accessed 27 February 2018).

Dorner, S.(1990) *Psychological problems*. in Neuropathic bladder in childhood. Clinics in Developmental Medicine 111 pp 96-110. London Mackeith Press

Doug, M., Adi, Y., Williams, J., Hall, M., Kelly, D., Petchy, R. (2011) Transitions to adult services for children and young people with palliative care needs, British Medical Journal Supportive and Palliative Care, Vol. 1., Issue 2.

Doug, M., Adi, Y., Williams, J., Hall, M., Kelly, D., Petchy, R. and Carter, Y.H. (2011) Transitions to adult services for children and young people with palliative care needs, online at Archives of Diseases of Childhood, Vol. 96, pp 78–84. DOI: 10.1136/adc.2009.163931.

Draper, J. (2000) 'Fathers in the making: Men, Bodies and Babies', Ph.D Thesis, *University of Hull*.

Draper, J. (2002) 'Fatherhood as transition: The contemporary relevance of transition theory', in Horrocks, C., Milnes, K., Roberts, B. and Robinson, D. (eds) *Narrative, Memory and Life Transitions*, Narrative and memory research group, Huddersfield, UK: University of Huddersfield Press, pp. 85–94.

Draper, J. (2003) 'Men's passage to fatherhood: an analysis of the contemporary relevance of transition theory', *Nursing Enquiry*, Vol. 10, No. 1, pp. 66–78.

Duckett, P. S. and Pratt, R. (2001) 'The researched opinions on research: Visually impaired people and visual impairment research', *Disability and Society*, vol. 16, no. 6, pp. 815–835.

Dynavox (AVdV) (n.d.) Dynavox [Online]. Available at <https://dynavox.com/index.php?route=product/search&search=dynavox%20comput> (Accessed 30 January 2018).

Earle, S. (2001) 'Disability, facilitated sex and the role of the nurse', *Journal of Advanced Nursing*, vol. 36, no. 3, pp. 433–440.

Earle, S. (2014) 'A life course approach to human reproduction', *Human Fertility*, vol. 17, no. 3, pp. 151–153.

Earle, S. and Church, S. (2004a) 'Disability and reproduction', *Practising Midwife* Vol. 7 No. 8, pp. 32–4.

Earle, S. and Church, S. (2004b) 'Sexuality and reproduction', *Practising Midwife* Vol. 7 No. 4, pp. 28–30.

Earle, S., Foley, P., Komaromory, C., and Lloyd, C.E. (2008) Conceptualising reproductive loss: a Social Sciences' perspective, *Human Fertility*, Vol. 11, No.4, pp. 259–62.

Earle, S., Komaromy, C., Foley, P. and Lloyd, C. (2007) 'The social dimensions of reproductive loss', *The Practising Midwife*, June, vol. 10, no. 6, pp. 28–34.

Earle, S., Komaromy, C. and Layne, L. (eds) (2012) *Understanding Reproductive Loss: Perspectives on Life, Death and Fertility*, Ashgate.

Earle, S. and Letherby, G. (2002) 'Whose choice is it anyway? Decision making, control and conception', *Human Fertility*, vol. 5, no. 2, pp. 39–41.

- Earle, S. and Letherby, G. (2007) 'Conceiving time? Women who do or do not conceive', *Sociology of Health and Illness*, vol. 29, no. 2, pp. 233–250.
- Earle, S. and Lloyd, C. E. (2012) 'Diabetes: A loss of reproductive normalcy?' in Earle, S., Komaromy, C. and Layne, L. (eds) *Understanding Reproductive Loss: International Perspectives on Life, Death and Fertility*, Ashgate.
- Earle, S., Tilley, L., Walmsley, J. and Atkinson, D. (2012) 'Sterilisation and women with learning disabilities: An international issue', in Earle, S., Komaromy, C. and Layne, L. (eds) *Understanding Reproductive Loss: Perspectives on Life, Death and Fertility*, Ashgate.
- Edenborough, F. P. and Morton, A. M. (2010) 'Cystic fibrosis – a guide for clinicians in reproductive and obstetric medicine', *Fetal and Maternal Medicine Review*, vol. 21, no. 1, pp. 36–54.
- Edwards, N. (2016) 'Women's reflections on formal sex education and the advantage of gaining informal sexual knowledge through a feminist lens', *Sex Education: Sexuality, Society and Learning*, Vol. 16, No. 3 pp. 266–78.
- Eiser, C. (1993) 'Growing Up with a Chronic Disease: The Impact on Children and Their Families', *Jessica Kingsley Publishers*.
- Elder, G. H., Jr. (1975) 'Age differentiation and the Life course', *Annual Review of Sociology*, Vol. 1, No. 1, pp. 165–90.
- Elder, G. H., Jr. (1978) 'Family history and the life course', in Hareven, T. K. (ed) *Transitions*, pp. 17–64, New York: Academic Press.

Elder, G. H., Jr. (1985) 'Life course dynamics: trajectories and transitions 1968-1980', *Ithaca New York, Cornell University Press*.

Elder, G. H., Jr. (1994) 'Time, human agency, and social change: Perspectives on the life course', *Social Psychology Quarterly*, vol. 57, no. 1, pp. 4–15.

Elder, G. H., Jr. (1995) 'The life course paradigm: social changes and individual development', in Moen, P., Elder, G. H., Jr. and Luscher, K. (eds) *Examining Lives in Context*, Washington, DC: American Psychological Association, pp. 101–139.

Elder, G.H., Jr. (1996) 'Human lives in changing societies', in Cairns, R. B., Elder. G. H. and Costello, E. J. (eds) *Developmental Science*, pp. 31–62, Cambridge University Press: Melbourne, Australia.

Elder, G. H., Jr. (1998) 'Life course and human development', in Damon, W. (ed) *Handbook of Child Psychology*, pp. 939–991, New York, Wiley.

Elder, G. H., Jr. (2000) 'The life course', in Borgatta, E. F. and Montgomery, R. J. V. (eds) *Encyclopedia of Sociology*, 2nd edn, vol. 3, pp. 1614–1622, New York, Macmillan.

Elder, G. H., Jr. and Johnson, M. K. (2003) 'The life course and human development: Challenges, lessons, and new directions', in Settersten, R. A. (ed) *Invitation to the Life Course: Toward New Understandings of Later Life*, Amityville, NY, Baywood, pp. 3–19.

Elder, G. H., Jr., Johnson, M. K. and Crosnoe, R. (2003) 'The emergence and development of life course theory', in Mortimer, J.T., Shanahan, N.J. (Eds) *Handbook of the life course*. Boston MA, Springer.

Ellis, E. (2015) *The Experiences of People with Learning Difficulties Living in a Rural Area, Doctoral thesis, Milton Keynes, The Open University.*

Ellis, E. (2017) '*Making decisions together? Exploring the decision-making process in an inclusive research project*', *Disability and Society* [Online]. DOI: 10.1080/09687599.2017.1392932 (Accessed 10 March 2018).

Evans, D. (1993) '*Sexual Citizenship: The Material Construction of Sexualities*', London, *Routledge.*

Family Planning Association (2015) '*Online sex and relationships education resource for primary schools*', available at www.fpa.org.uk/schools-and-teachers/online-sre-and-pshe-primary-schools?ems= (Accessed 7 October 2015).

Faugier, J. and Sargeant, M. (1997) '*Sampling hard to reach populations*', *Journal of Advanced Nursing*, vol. 26, no. 4, pp. 790–797.

Forbat, L. and Kelly (2012) '*Discussing the sexual consequences of treatment in radiotherapy and urology consultations with couples affected by prostate cancer*', *British Journal of Urology*, vol. 109, no. 1, pp. 98–103.

Foreman, N. (2018) '*Juvenile Pilocytic Astrocytoma*', *NORD* [Online]. Available online at <https://rarediseases.org/rare-diseases/juvenile-pilocytic-astrocytoma/> (Accessed 5 April 2018).

Fraser, L. K., Jarvis, S. W., Moran, N. E., Aldridge, J., Parslow, R. C. and Beresford, B. A. (2015) *Children in Scotland Requiring Palliative Care: Identifying Numbers and Needs*, The ChiSP Study, University of York.

Fraser, L. K. and Norman, P. (2017) 'The use of routine data in health research: an example from palliative care', *SAGE Research Methods Cases*, SAGE [Online]. DOI: [10.4135/9781526423658](https://doi.org/10.4135/9781526423658) (Accessed 11 March 2018).

Fraser, L. K., Lidstone, V., Miller, M., Aldridge, J., Norman, P., McKinney, P. A. and Parslow, R. C. (2014) 'Patterns of diagnoses among children and young adults with life-limiting conditions: A secondary analysis of a national dataset', *Palliative Medicine*, vol. 28, no. 6, pp. 513–520 [Online]. DOI: [10.1177/0269216314528743](https://doi.org/10.1177/0269216314528743) (Accessed 11 March 2018).

Fraser, L. K., Aldridge, L., Manning, S., O'Leary, S., Miller, M., McCulloch, R. and Childs, A. M. (2011) 'Hospice provision and usage amongst young people with neuromuscular disease in the United Kingdom', *European Journal of Paediatric Neurology*, vol. 15, no. 4, pp. 326–330 [Online]. DOI: [10.1016/j.ejpn.2011.02.003](https://doi.org/10.1016/j.ejpn.2011.02.003) (Accessed 11 March 2018).

Fraser, L. K., Miller, M., Aldridge, J., McKinney, P. A. and Parslow, R. C. (2013) 'Prevalence of life-limiting and life-threatening conditions in young adults in England 2000–2010', *Final Report for Together for Short Lives*, York, Department of Health Sciences, University of York. Available from [http://www.togetherforshortlives.org.uk/assets/0000/6736/TOGETHER FOR SHORT LIVESAdultReport2013Final.pdf](http://www.togetherforshortlives.org.uk/assets/0000/6736/TOGETHER_FOR_SHORT_LIVESAdultReport2013Final.pdf) (Accessed 11 March 2018).

Fraser, L. K., Miller, M., Hain, R., Norman, P., Aldridge, J., McKinney, P. A. and Parslow, R. C. (2012) 'Rising national prevalence of life-limiting conditions in children in England', *Pediatrics*, vol. 129, no. 4, pp. e923–e929 [Online]. DOI: [10.1542/peds.2011-2846](https://doi.org/10.1542/peds.2011-2846) (Accessed 6 October 2015).

Frayman, K. and Sawyer, S. M. (2015) 'Sexual and reproductive health in cystic fibrosis: A lifecourse perspective', *Lancet Respiratory Medicine*, vol. 3, no. 1, pp. 70–86

Froggatt, K. (1997) 'Signposts on the journey: the place of ritual in spiritual care'. *International Journal of Palliative Nursing* 3(1), 42– 46.

Garbutt, R. (2010) 'Exploring the barriers to sex and relationships for people with learning difficulties', in Shuttleworth, R. and Sanders, T. (eds) *Sex and Disability: Politics, Identity, and Access*, Leeds, The Disability Press.

Gawande, A. (2015) *Being Mortal: Illness, Medicine and What Matters in the End*, Metropolitan Books, Wellcome Collection.

Gee, L., Abbott, J., Conway, SP. Etherington, C., Webb, AK (2003) 'Quality of life in cystic fibrosis, general health perceptions and disease severity. *Journal of Cystic Fibrosis*', Elsevier. [https://doi.org/10.1016/S1569-1993\(03\)00093-6](https://doi.org/10.1016/S1569-1993(03)00093-6) (Accessed 17 February 2019)

General Data Protection Regulation (2018) available at: www.ibm.com/security/data-security, accessed in 4 April 2018.

Genetic Disorders UK (2018) 'About Genetic Disorders' [Online]. Available at <http://www.geneticdisordersuk.org/aboutgeneticdisorders> (Accessed 16 February 2018).

Giele, J.Z. and Elder, G.H. (1998) 'Methods of Life Course Research: Qualitative and Quantitative Approaches'. *Sage: Thousand Oaks, CA*. [Online]. DOI: 10.4135/9781483348919 (Accessed 2 February 2018)

Giele, J. Z.(2013) 'Innovation in the typical life course', in Giele, J. Z. and Elder, G. H., Jr. (eds) *Methods of Life Course Research: Qualitative and Quantitative Approaches*, pp. 231–263, Sage, Thousand Oaks, CA.

Gill, R. (2008) 'Empowerment/sexism: Figuring female sexual agency in contemporary advertising', *Feminism and Psychology*, vol. 18, no. 1, pp. 35–60 [Online]. DOI: 10.1177/0959353507084950 (Accessed 29 March 2018).

Gillespie-Sells, K., Hill, M., and Robbins, B. (1998) 'She Dances to Different Drums'. London, *King's Fund*.

Glaser, B. and Strauss, A. (1967). 'The discovery of grounded theory: strategies for qualitative research', *Weidenfeld and Nicolson*, London.

Goldman, A. (1994) 'Care of the dying child', *Oxford Medical Publications*. (Accessed 12 April 2018)

Goldman, A. (2018) 'A personal perspective of palliative care for children from the UK', ChiPPS Children's Project on Palliative/Hospice Services, in 'On celebrating the past and the future of pediatric palliative/hospice care', *ChiPPS and the ChiPPS E-Journal*, 50th issue. Released in collaboration with the National Hospice and Palliative Care Organization.

Goldman, A., Hain, R. and Liben, S. (eds) (2012) *The Oxford Textbook of Palliative Care for Children*, 2nd Edn, Oxford: Oxford University Press.

Gomm, R., Needham, G. and Bullman, A. (2001) *Evaluating Research in Health and Social Care*, The Open University, Sage Publications.

Goodley, D., Liddiard, K. and Runswick-Cole K. (2017) 'Feeling disability: theories of affect and critical disability studies', *Disability and Society*, vol. 33, no. 2, pp. 197–217.

Governance Arrangements for Research Ethics Committees (GAfREC) (2012) Available at www.nres.nhs.uk/NRES Publications (Accessed 14 April 2018).

Graham, R., Embleton, N., Farnworth, A., Mason, K., Rankin, R., and Robson, S. (2012). 'Importance of Professional Discretion in Caring for a Patient Group with Diverse Views' in ' *Understanding reproductive loss*. eds. Earle, S., Komaromy, C. and Layne, L. Ashgate.

Gravelle, A. (1997) 'Caring for a child with a progressive illness during the complex phase: parents' experience of facing adversity'. *Journal of Advanced Nursing* 25(4), 738– 745.

Greco, M. and Stenner, P. (2017) 'From paradox to pattern shift: Conceptualising liminal hotspots and their affective dynamics', *Theory and Psychology*, vol. 27, no. 2, pp. 147–166.

Griffith, R. and Tegenah, C. (2014) 'Assessing capacity to consent to sexual relations: a guide for nurses', *British Journal of Community Nursing*, April, vol. 18, no. 4, pp. 198–201.

Grob, A., Krings, F. and Bangerter, A. (2001) 'Life markers in biographical narratives of people from three cohorts: a life span perspective in historical context', *Human Development*, vol. 44, no. 4, pp. 171–90.

The Guardian, 2017, *Joanna Morhead*, 29 April 2017. On line at <https://www.theguardian.com/uk> (Accessed 7 December 2017)

Hagestad, G. O. (2003) 'Interdependent lives and relationships in changing times: a life course view of families and aging', in Settersten, R.A. and Hendricks, J. (Eds)

Invitation to the life course: towards new understandings of later life. *Amityville, New York: Baywood*, pp. 135–39.

Hain, R. and Devins, M. (2011) Directory of Life-Limiting Conditions. Available at: http://www.togetherforshortlives.org.uk/assets/0000/7089/Directory_of_LLC_v1.3.pdf [accessed on 22/3/2017].

Hain, R., Davies, I., Harrup, R. and Torshkav (2013a) 'Development and utility of a dictionary to establish definitions, incidence and prevalence of life-limiting conditions in children' (www.togetherforshortlives.org.uk).

Hain, R., Devins, M., Hastings, R. and Noyes, J. (2013b) 'Paediatric palliative care: development and pilot study of a "Directory" of life-limiting conditions', *BMC Palliative Care*, vol. 12, no. 1, p. 43 [Online]. DOI: 10.1186/1472-684X-12-43 (Accessed March 11 2018).

Hammersley, M., (2007) 'The issue of quality in qualitative research', *International Journal of Research & Method in Education*, 30:3, p287-305

Hammersley, M. (2010) 'Reproducing or constructing? Some questions about transcription in social research', *Qualitative Research*, Vol 10(5) 553-569.

Hammersley, M. (2013) 'Pragmatism, Paradigms, and Research as Reflective Practice,' key note presentation, May, The Open University/University of Greenwich, School of Education.

Hammersley, M. (2014)' What is Qualitative Research'? London: Bloomsbury Academic.

Hammersley, M. (2015) 'Sampling and thematic analysis: a response to Fugard and Potts'.

Hammersley, M. and Gomm, R. (1997) 'Bias in social research', *Sociological Research Online*, vol. 2, no. 1 [Online]. Available at <http://www.socresonline.org.uk/2/1/2.html> (Accessed 28 November 2017).

Hanson, J. (2013) 'Education developers as researchers: the contribution of insider research to enhancing understanding of role, identity and practice', *Innovations in Education and Teaching International*, vol. 50, no. 4, pp. 388–398.

Harris, N. (2018) 'Research data capture repositories and children's palliative care', *Paper for the National Joint Research Group, Association for Paediatric Palliative Care Medicine and Together for Short Lives*, March 2017.

Helen and Douglas House (2014) *Transition and Beyond Toolkit: For Professionals Working with Young People Who Have Palliative Care Needs*.

Help the Hospices (2013) 'Commission into the Future of Hospice Care/Hospice UK: The Role of Hospices in Supporting Young People in Transition from Children to Adult Services', www.hospiceuk.org. (Accessed 15 February 2016).

Hockey, J. and James, A. (1993) 'Growing Up and Growing Older: Ageing and Dependency in the Life Course'. *Sage: London*.

Hodge A. (1995) Addressing issues of sexuality with spinal cord injured persons. *Orthopaedic Nursing*, 14(3), 21-24.

Holland, J. and Ramazanoglu, C. (1994) 'Coming to conclusions: Power and interpretation in researching young women's sexuality', in Maynard, M. and Purvis, J. (eds) *Researching Women's Lives*, London and Bristol, Taylor and Francis.

Hollomotz, A. (2010) 'Sexual "vulnerability" of people with learning difficulties: A self-fulfilling prophecy', in Shuttleworth, R. and Sanders, T. (eds) *Sex and Disability: Politics, Identity, and Access*, Leeds, The Disability Press.

Hordern, A. J. and Currow, D. (2003) 'A patient-centred approach to sexuality in the face of life-limiting illness', *The Medical Journal of Australia*, vol. 179, no. 6, S8–S11.

Hordern, A. J. and Street, A. F. (2007a) 'Constructions of sexuality and intimacy after cancer: patient and health professional perspectives', *Social Science and Medicine*, vol. 64, no. 8, pp. 1704–1718.

Hordern, A. J. and Street, A. F. (2007b) 'Issues of intimacy and sexuality in the face of cancer: The patient perspective', *Cancer Nursing*, vol. 30, no. 6, November/December, pp. E11–E18.

Hordern, A. J. and Street, A. F. (2007c) 'Communicating about patient sexuality and intimacy after cancer: Mismatched expectations and unmet needs', *The Medical Journal of Australia*, vol. 186, no. 5, pp. 224–227

Hordern, A. J. and Street, A. F. (2007d) 'Let's talk about sex: Risky business for cancer and palliative care clinicians', *Contemporary Nurse*, vol. 27, no. 1, pp. 49–60.

Hospice UK (2018) *Highlights 2017-2018*: www.hospiceuk.org

Howlett, C., Swain, M., Fitzmaurice, N., Mountford, K. and Love, P. (1997) 'Sexuality: The neglected component in palliative care', *International Journal of Palliative Nursing*, vol. 3, no. 4, pp. 218–221.

Hughes-Hallet, T., Craft, A. and Davies, C. (2011) 'Palliative Care Funding Review: Funding the Right Care and Support for Everyone', *Department of Health, London*.

Human Fertilisation and Embryology Authority (HFEA) (n.d.) HEFA website welcome [Online]. Available at <https://www.hfea.gov.uk/> (Accessed 1 April 2018).

Human Rights Act (1998) 'Chapter 42' [Online]. Available at <http://www.legislation.gov.uk/ukpga/1998/42/data.pdf> (Accessed 1 April 2018).

Hunt, J. A., Swallow, V. and Twycross, A. (2014) 'A peer-driven community-based doctoral supervisory model: Development from an evaluation of an ethics workshop for healthcare professionals undertaking research with children', *Health Science Journal*, vol. 8, no. 2, pp. 154–165.

IBM (2018) *General Data Protection Regulation (GDPR)*, IBM Security [Online]. Available at <https://www.ibm.com/security/data-security/gdpr> (Accessed 2 April 2018).

Jackson, S. (1999) *Heterosexuality in Question*, London, Sage Publications Ltd.

Jepson, M., Abbott, D. W. F. and Hastie, J. (2015) "'This is another personal question": Research interviews and discussing sensitive issues with men with life-limiting conditions', *International Journal of Men's Health*, vol. 14, no. 3, pp. 273–286.

Jordan, J., Price, J. and Prior, L. (2015) 'Disorders and disconnections: parents' experiences of liminality when caring for their dying child', *Sociology of Health and Illness*, Vol 37, No. 6. Pp. 839–55.

Karlsson M., Friberg, F., Wallengren, C. and Ohlen, J. (2014) 'Meanings of existential uncertainty for people diagnosed with cancer and receiving palliative treatment: a life-world phenomenological study', *British Medical Care*, Vol. 13, No. 28 pp. 1–9.

Kelly, D. M. (2009) 'Changed men: the embodied impact of prostate cancer', *Qualitative Health Research*, vol. 19, no. 2, pp. 151–163 [Online]. DOI: [10.1177/1049732308328067](https://doi.org/10.1177/1049732308328067) (Accessed November 3 2017).

Kelly, D. M. (2012) 'Palliative care for adolescents and young adults', in Faull, C. et al. (eds) *Handbook of Palliative Care*, Blackwell, pp. 247–256.

Kelly, D. M. (2013) 'Developing age appropriate psychosexual support for adolescent cancer survivors: A discussion paper', *Journal of Sexual Medicine*, vol. 10, no. S1, pp. 133–138 [Online]. DOI: [10.1111/jsm.12048](https://doi.org/10.1111/jsm.12048) (Accessed 13 November 2017).

Kelly, D., Forbat, L., Marshall-Lucette, S., White, I (2015). 'Co-constructing sexual recovery after prostate cancer: a qualitative study with couples'. *Translational andrology and urology* 4(2): 131.

Kelly, D. M. and Hooker, L. 2007. 'Evidence based cancer policy: the needs of teenagers and young adults' [Editorial]. *European Journal of Oncology Nursing* 11(1), pp. 4-5. ([10.1016/j.ejon.2006.12.004](https://doi.org/10.1016/j.ejon.2006.12.004))

Kelly, D. M., and Vougioukalou, S. (2017) 'The impact of cancer treatment on sexuality and relationships for teenage and young adult cancer survivors', in Reisman, Y. and

Gianotten, W. L. (eds) *Cancer, Intimacy and Sexuality: A Practical Approach*, Springer, pp. 249–256.

Kimmel M. (2018) Healthy Sexuality, available at:
<https://sociologyproject110.weebly.com/what-is-sexual-agency.html>, accessed 7 April 2018.

Kinsella, E.A. (2006) 'Constructivist underpinning in Donald Schon's theory of reflective practice: echoes of Nelson Goodman', *Reflective Practice*, Vol. 7, No. 3, pp277–86.

Kitzinger, C. and Kitzinger, S. (2007) 'Birth trauma: talking with women and the value of conversational analysis', *British Journal of Midwifery*, Vol. 15, pp. 260–64.

Komaromy, C., Earle, S., Foley, P. and Lloyd, C. (2007) 'Reproductive loss and the moment of death', *The Practising Midwife*.

Kralik, D. (2002) The quest for ordinariness: transition experienced by midlife women living with chronic illness. *Journal of Advanced Nursing* 39(2), 146– 154.

Kralik, D., Visentin, K. and Van Loon, A. (2006), 'Transition: a literature review. *Journal of Advanced Nursing*, 55: 320-329. doi:[10.1111/j.1365-2648.2006.03899.x](https://doi.org/10.1111/j.1365-2648.2006.03899.x) (Accessed 16 February 2019)

Livescribe digital pen; see <https://www.livescribe.com/site/> On line (Accessed 23 November 2013).

Law, J. and Urry, J. (2003) 'Enacting the Social'. Published by the Department of Sociology and the Centre for Science Studies. *Lancaster University, UK*.

<http://www.lancaster.ac.uk/fass/resources/sociology-online-papers/papers/law-urry-enacting-the-social.pdf> Accessed 4 February 2019.

Ledger, S., Earle, S., Tilley, E. and Walmsley, J. (2016) 'Contraceptive decision-making and women with learning disabilities', *Sexualities*, vol. 19, no. 5–6, pp. 698–724.

Ledger, S., Shufflebotham L., and Walmsley, J. (2016) Disability, in Denny, E., Earle, S., and Hewison, A. (Eds) *Sociology for Nurses 3rd Edition*, Cambridge: Polity Press.

Lee, R. (1993) *Doing Research on Sensitive Topics*, University of London, UK, Sage Publications.

Lee, D. J. and Stvilia, B. (2017) 'Practices of research data curation in institutional repositories: A qualitative view from repository staff', *PLoS ONE*, Public Library of Science, vol. 12, no. 3.

Letherby G. (2010) 'The Changing Life Course in British Sociology'. In: Burnett J., Jeffers S., Thomas G. (eds) *New Social Connections*. *Palgrave Macmillan*, London

Levi, R. (2006) 'Issues of Sexuality Amongst Adolescents with Life-limiting Conditions', *ChiPPS Pediatric Palliative Care Newsletter*, no. 5, September.

Levy, R. and Bühlmann, F. (2016) 'Towards a socio-structural framework for life course analysis', *Advances in Life Course Research*, vol. 30, pp. 30–42 [Online]. DOI: [org/10.1016/j.alcr.2016.03.005](https://doi.org/10.1016/j.alcr.2016.03.005) (Accessed 14 March 2018).

Liddiard, K. (2011) '(S)exploring Disability: Intimacies, Sexualities and Disabilities', PhD Doctor of Philosophy, *Warwick University*.

Liddiard, K. (2014b) “‘I never felt like she was just doing it for the money’”: Disabled men’s intimate (gendered) realities of purchasing sexual pleasure and intimacy’, *Sexualities*, vol. 17, no. 7, pp. 837–855.

Liddiard, K. (2015) ‘The work of disabled identities in intimate relationships’, in Zinn, M. B. et al. (eds) *Gender Through the Prism of Difference*, Oxford University Press, USA
Liddiard, K. (2014a) ‘The work of disabled identities in intimate relationships’, *Disability and Society*, vol. 29, no. 1, pp. 115–128.

Liddiard, K. (2018) ‘The Intimate Lives of Disabled People’, *Routledge, Taylor Francis*.

Liddiard, K. and Goodley, D. (2015) ‘Disability and impairment’, in *The Wiley-Blackwell Encyclopedia of Social Theory*, West Sussex, John Wiley and Sons.

Lincoln, Y.S. and Guba, E.G. (1985) ‘Naturalistic Enquiry’, *Sage*: Beverly Hills, CA.

Lund, E. M. and Johnson B. A. (2015) ‘Asexuality and disability: Strange but compatible bedfellows’, *Sexuality and Disability*, vol. 33, no. 1, pp. 123–132, Springer Science, Business Media New York [Online]. DOI: 10.1007/s11195-014-9378-0 (Accessed 11 October 2017).

MacLeod, A., Lewis, A. and Robertson, S. (2013) ‘CHARLIE: PLEASE RESPOND!’ Using a participatory methodology with individuals on the autism spectrum. *International Journal of Research and Method in Education* 37 (4), 407-420.
DOI.org/10.1080/1743727X.2013.776528

MacMillan, R. and Copher, R. (2005) ‘Families in the life course: independency of roles, role configuration, and pathways’, *Journal of Marriage and Family*, 67, pp. 858–79.

Magnusson, D. and Torestad, B. (1993) 'A holistic view of personality: a model revisited', *Annual Review of Psychology*, Vol. 44, No.1, pp. 427–52.

Makaton (2015) 'Sex Education Symbols Book' [Online]. Available at www.makaton.org/shop/shopping/stockDetails/Sex-Education-Symbols-Book (Accessed on 28 February 2018).

Manhas, K. P., Page, S., Dodd, S. X., Letourneau, N., Ambrose, A. and Cui, X. (2015) 'Parent perspectives on privacy and governance for a pediatric repository of non-biological, research data', *Journal of Empirical Research on Human Research Ethics*, SAGE Publications, Sage CA: Los Angeles, CA, vol. 10, no. 1, pp. 88–99.

Marie Curie Cancer Care and Together for Short Lives (2012) 'Don't Let Me Down: Ensuring a Good Transition for Young People with Palliative Care Needs' at www.mariecurie.org.uk

Marshall, V. and Mueller, M. (2003) Theoretical roots of the life course perspective, in Heinz, W. and Marhsall, V. (Eds) *Social dynamics of the life course*, New York: De Gruiter, pp. 3–32.

Marston, J. and Chambers, L. (2012) 'International aspects', in Goldman, A., Hain, R. and Liben, S. (eds) *Oxford Textbook of Palliative Care for Children*, pp. 458–470.

Maudsley, D.B. (1979) 'A Theory of Meta-Learning and Principles of Facilitation: An Organismic Perspective'. University of Toronto, 1979. (40, 8,4354-4355-A)

McCabe, M. P. (1999) 'Sexual knowledge, experience and feelings of people living with disability', *Sexuality and Disability*, Vol 17. No. 2 pp. 157–70.

McCall, L. (2005) 'The complexity of intersectionality', *Signs: Journal of Women in Culture and Society*, vol. 30, pp. 1771–1800 [Online]. Available at <https://www.nhs.uk/conditions/contraception/contraceptive-injection/> (Accessed 16 February 2018).

McFadyen, J. and Rankin, J. (2016) The roll of gate-keepers in research: learning from reflexivity and reflection, *Journal of Nursing and Health Care*, Vol. 4, No.1, pp 82–88.

McNamara-Goodger, K. and Feudtner, C. (2012) 'History and epidemiology', in Goldman, A., Hain, R. and Liben, S. (eds) *Oxford Textbook of Palliative Care for Children*, pp. 3–12.

McNeill, P. (1992) 'Research Methods' *Society Now*, 2nd edn, pp. 10–12, Routledge.

Metcalfe, A., Plumridge, G., Coad, J., Shanks, A. and Gill, P. (2011) 'Parents' and children's communication about genetic risk: a qualitative study, learning from families' experiences', *European Journal of Human Genetics*, vol. 19, no. 6, pp. 640–646 [Online]. DOI: 10.1038/ejhg.2010.258 (Accessed 3 March 2018).

Milligan, M. and Neudfeldt, A. (2001) The myth of asexuality: a survey of social and empirical evidence, *Sexuality and Disability*, Vol. 4, pp. 91–109.

Mitchell, W. A., Beresford, B. A., Brooks, J., Moran, N. E. and Glendinning, C. (2016) 'Taking on choice and control in personal care and support: The experiences of physically disabled young adults', *Journal of Social Work*, May, vol. 17, no. 4.

Mitchell, K. and Wellings, K. (1998) *Talking About Sexual Health: Interviews with Young People and Professionals*, Project Report, Health Education Authority, London.

Morgan, D. J. R., Blackburn, M. C. and Bax, M. C. O. (1995) 'Setting up new medical services: Adults with spina bifida and/or hydrocephalus', , vol. 71, no. 831, pp. 17-21, *Postgraduate Medical Journal*, The Fellowship of Postgraduate Medicine.

Morrow, V., Boddy, J. and Lamb, R. (2014) 'The Ethics of Secondary Data Analysis: Learning from the Experience of Sharing Qualitative Data from Young People and their Families in an International Study of Childhood Poverty', *National Centre for Research Methods*.

Muckaden, M., Dighe M., Balaji, P. D., Dhiliwal, S., Tilve, P. P. Jadhav, S. and Goswani, S. (2011) 'Paediatric palliative care: Theory to practice', *Indian Journal of Palliative Care*, vol. 17, pp. S52–S60 [Online] DOI: 10.4103/0973-1075.76244 (Accessed 29 March 2018).

Navon, L and Amira, M. (2004) 'Liminality as biographical disruption: unclassifiability following hormonal therapy for advanced prostate cancer'. *Social Science & Medicine* (1982) 58, 2337– 2347.

Newton, V. (2016) 'It's good to be able to talk': An exploration of the complexities of participant and researcher relationships when conducting sensitive research', *Women's Studies International Forum*, March–April, vol. 61, pp. 93–99.

NHS Inform (2019) Available at www.nhsinform.scot/illnesses-and-conditions/sexual-and-reproductive/loss-of-libido (Accessed 19 February 2019).

NHS England (2017) 'The NHS is its people' [Online]. Available at www.nhsengland.co.uk (Accessed 18 April 2018).

Neinstein, L. (2016) 'Neinstein's Adolescent and Young Adult Health Care: A Practical Guide (Adolescent Health Care a Practical Guide)' Sixth Edition. Wolters Kluwer. ISBN-13: 978-1451190083

Norman, P. and Fraser, L. (2014) 'Prevalence of life-limiting conditions in children and young people in England: Time trends by area type', *Health and Place*, vol. 26, pp. 171–179 [Online]. DOI: [10.1016/j.healthplace.2014.01.002](https://doi.org/10.1016/j.healthplace.2014.01.002) (Accessed 11 January 2018).

Nursing and Midwifery Council. (2018). *The code: Professional standards of practice and behaviour for nurses, midwives and nursing associates*. Retrieved from <http://www.nmc.org.uk/globalassets/sitedocuments/nmc-publications/revised-new-nmc-code.pdf> (Accessed 18th March 2018)

NVIVO Qualitative Software. Retrieved from <http://www.qsrinternational.com/nvivo/what-is-nvivo>. (Accessed 17 March 2018)

Office for National Statistics (online) www.ons.gov.uk accessed 3 April 2018.

Oliver, M. (2004) 'The social model in action', in Barnes, C. and Mercer, G. (eds) *Implementing the Social Model of Disability: Theory and Research*, Leeds, The Disability Press, pp. 18–31.

Oliveira, D.L. (2011) 'The use of focus groups to investigate sensitive topics: an example taken from research on adolescent girls' perceptions about sexual risks'. *Ciência & Saúde Coletiva* 16 (7), 3093-102.

The Open University Human Research Ethics Committee (2014) 'Ethics Principles for Research Involving Human Participants' [Online]. Available at

<http://www.open.ac.uk/research/sites/www.open.ac.uk.research/files/files/ecms/research-pr/web-content/Ethics-Principles-for-Research-involving-Human-Participants.pdf> (Accessed 2 February 2018).

The Open University with Together for Short Lives (2016) 'Talking About Sex, Sexuality and Relationships; Guidance and Standards for those Working with Young People with Life-limiting or Life-threatening Conditions' [Online]. Available at <https://www.togetherforshortlives.org.uk/resource/talking-sex-sexuality-relationships-guidance-standards/> (Accessed 26 April 2018).

O'Rand A. M. (1996) 'Precious and the precocious: understanding cumulative disadvantage and cumulative advantage over the life course', *Gerontology*, Vol. 36,2 pp. 230–38.

Parsons, T. (1951) 'The social system'. London: *Routledge and Kegan-Paul*.

Penrod, J. (2007) 'Living with uncertainty: concept advancement', Wiley Online Library, (Accessed 3 March 2018).

Peou, C. and Zinn J. O. (2013) 'Managing Uncertainties of the Life Course in Transformative Society – Experiences of a New Generation of Young Cambodians', *School of Social and Political Sciences, University of Melbourne*.

Petroni, S., Madhumita, D and Sawyer, S.M (2019). 'Protection versus rights: age of marriage versus age of sexual consent'. *The Lancet Child & Adolescent Health* 3(4): 274-280.

Philp, M. and Duckworth, D. (1982). 'Children with disabilities and their families: a review of research'. NFER-Nelson ; Atlantic Highlands, N.J. : Distributed in the USA by *Humanities Press, Windsor, Berks.*

Pilcher, J (2005). 'School sex education: policy and practice in England 1870 to 2000, *Sex Education*, 5:2, 153-170, DOI: 10.1080/14681810500038848

Plummer, K. (1995) 'Telling Sexual Stories: Power, Change and Social Worlds', London, *Routledge.*

Plummer, K. (2003) 'Intimate Citizenship: Private Decisions and Public Dialogues', *Seattle, WA.*

Plummer, K. (2005) 'Intimate citizenship in an unjust world', in Romero, M. and Margolis, E. (eds) *Wiley Online Library* [Online]. Available at <https://doi.org/10.1002/9780470996973.ch5> (Accessed 13 April 2018).

Przybylo, E. (2013) 'Producing facts: empirical asexuality and the scientific study of sex', *Feminism and Psychology*, vol. 23, pp. 224–242 [Online]. DOI: 10.1177/0959353512443668 (Accessed 25 April 2018).

Punch, K. (2008) 'Introduction to Social Research: Quantitative and Qualitative' Approaches, 2nd edn, Sage.

Ramazanoglu, C. and Holland, J. (2002) 'Part III: Meeting challenges, making choices', in *Feminist Methodology: Challenges and Choices*, pp. 152–154, Thousand Oaks, CA: SAGE Publications.

Randall, F. and Downie, R. S. (2010) 'End of Life Choices', *Oxford University Press.*

Rare Diseases UK (2018) available at www.raredisease.org.uk (accessed 10 February 2018).

Read, A. P. and Donnai, D. (2011a) 'New Clinical Genetics', 2nd edn, *Scion, Banbury*.

Read, A. P. and Donnai, D. (2012) 'What can be offered to couples at (possibly) increased genetic risk?', *Journal of Community Genetics*, vol. 3, no. 3, pp. 167–174 [Online]. DOI: 10.1007/s12687-012-0105-1 (Accessed 10 March 2018).

Reeve, D. (2004) 'Psycho-emotional dimensions of disability and the social model', in Barnes, C. and Mercer, G. (eds) *Implementing the Social Model of Disability: Theory and Research*, Leeds, The Disability Press.

Renzetti, C.M. and Lee, R.M. (1993) *Researching sensitive topics*, Newbury Park, CA: Sage Publications.

Reiss, M. (1998) *The history of school sex education*, *Muslim Education Quarterly*, 15, 4–13.

Richardson, D. (1998) 'Sexuality and citizenship', *Sociology*, vol. 32, no. 1, pp. 83–100.

Richardson, D. (2000) 'Rethinking Sexuality', *London, SAGE*.

Richardson, D. (2004) 'Locating sexualities: From here to normality', *Sexualities*, vol. 7, no. 4, pp. 391–411.

Richardson, D. (2005) 'Desiring sameness? The rise of a neoliberal politics of normalisation', *Antipode*, vol. 37, no. 3, pp. 515–553.

Richardson, D. (2007) 'Patterned fluidities: (Re)imagining the relationship between gender and sexuality', *Sociology*, vol. 41, no. 3, pp. 457–474. Richardson, D. (2015) 'Rethinking sexual citizenship', *Sociology*, vol. 51, no. 2, pp. 208–224.

Richardson, D. (2017) 'Rethinking Sexual Citizenship. Sociology' *The Journal of the British Sociological Association* **51**(2): 208-224.

<https://doi.org/10.1177/0038038515609024> (Accessed March 3, 2018)

Royal College of Nursing (2014) 'Core knowledge: Sexual health for non-experts', *CPD Online Learning* [Online]. Available at www.rcn.org.uk/development/practice/cpd_online_learning/sexual_health_for_non-experts/core_knowledge (Accessed 6 October 2015).

Sabsay, L. (2013) 'Citizenship in the twilight zone? Sex work, the regulation of belonging and sexual democratization in Argentina', in Roseneil, S. (ed) *Beyond Citizenship? Feminism and the Transformation of Belonging*, Basingstoke, Palgrave Macmillan, pp. 160–183.

Sarrel L. J. and Sarrel P.M. (1981) 'Sexual unfolding' *Journal of Adolescent Health Care* **2**(2): 93–9. DOI: 10.1016/S0197-0070(81)80097-6.

Saunders, M.N.K. (2006) 'Gatekeeping, Sage Dictionary of Social Research Methods' (online), (Accessed 4 November 2017}.

Saunders, N. (2006) 'Gatekeepers'. In V. Jupp (Ed.), *SAGE dictionary of social research methods*. London: Sage. (Accessed March 19 2018).

Savage, D. (2011)' 'A Review of Palliative Care for Children and Young Adults in the Thames Valley', *Helen and Douglas House*.

Sawyer, S. M., Afifi, R. A., Bearinger, L. H., Blakemore, S. J., Dick, B., Ezay, A. and Patton, G. C. (2012) 'Adolescence: a foundation for future health', *The Lancet*, vol. 379, no. 9826, pp. 1630–1640.

Sawyer, S. M., Ambresin, A. E., Bennett, K. E. and Patton, G. C. (2014) 'A measurement framework for quality healthcare for adolescents in hospitals', *Journal of Adolescent Health*, vol. 55, no. 4, pp. 484–90.

Sawyer, S. M., Azzopardi, P. S., Wickremarathne, D. and Patton, G. C. (2018) 'The age of adolescence', *The Lancet Child and Adolescent Health*, vol. 2, no. 3, pp. 223–28.

Schön, D. (1983) *The Reflective Practitioner: How Professionals Think in Action*, New York, Basic Books.

Schön, D. (1987) 'Educating the Reflective Practitioner, Towards a New Design for Teaching and Learning in the Professions', *San Francisco*,

Jossey-Bass. Schwarze, M. L. and Taylor, L. J. (2017) 'Managing uncertainty — harnessing the power of scenario planning', *New England Journal of Medicine*, vol. 377, pp. 206–208.

Schostak, J. (2011) 'Interviewing and focus groups', in Somekh, B. and Lewin, C. (eds) *Theory and Methods in Social Research*, 2nd edn, London, UK: Sage Publications Ltd., pp. 61–68.

Settersten, Jr., R. A., Ottusch, T. M., and Schneider, B. (2005) *Becoming Adult: Meanings of Markers to Adulthood: An Essay* [Online]. Available at <http://www.ricksettersten.com> (Accessed 11 March 2011).

The Sexual Health Respect Toolkit (2018) [Online]. Available at <http://www.sexualrespect.com/> (Accessed 20 April 2018).

Shakespeare, T. (1996) 'Power and prejudice: Issues of gender, sexuality and disability', in Barton, L. (ed) *Disability and Society: Emerging Issues and Insights*, Harlow, Longman.

Shakespeare, T. (1999a) 'The sexual politics of disabled masculinity', *Sex and Disability*, vol. 17, no. 1, pp. 53–64.

Shakespeare, T. (1999b) "'Losing the plot"? Medical and activist discourses of contemporary genetics and disability', *Sociology of Health and Illness*, vol. 21, no. 5, pp. 669–68.

Shakespeare, T. (2000) 'Disabled sexuality: Toward rights and recognition', *Sexuality and Disability*, vol. 18, no. 3, pp. 159–166.

Shakespeare, T., Gillespie-Sells, K. and Davies, D. (1996) *Untold Desires: The Sexual Politics of Disability*, London and New York, Cassell.

Silver, S. and Lewins, A. 'Using Software in Qualitative Research: A Step-by-Step Guide'. Sage, Second Edition. ISBN 978-1-4462-4973-4.

Shuttleworth, R. P. (2010) 'The search for sexual intimacy for men with cerebral palsy', *Sexuality and Disability*, vol. 18, no. 4, pp. 263–281.

Sidebotham, P. and Appleton, J. V. (2014) 'From 2014 to 2015 and beyond: Using evidence to promote the protection of children worldwide', *Child Abuse Review*, vol. 23, no. 1, pp. 1–4 [Online]. DOI: [10.1002/car.2314](https://doi.org/10.1002/car.2314) (Accessed 12 April 2018).

Siebers, T. (2008) *Disability Theory*, Ann Arbor, MI, University of Michigan Press.

Silverman, D. (2004) *Qualitative Research: Theory, Method and Practice*, SAGE Publications. Sir James Munby, RE MM, 2007, EWHC, 2003.

Smart, J. F. (2009) *Disability, Society, and the Individual*, 2nd edn, Pro Ed, Austin.

Smith, J. Swallow, V., Coyne, I. (2015) 'Fathers' contributions to the management of their child's long-term medical condition: a narrative review of the literature'. *Journal of Pediatric Nursing: Nursing Care of Children and Families*.

Stein A. F., G. C., Wooley, T. C. and Baum, J. D. (1989) 'Life-threatening illness and hospice care,' *Archives of Disease in Childhood*, vol. 64, no. 5, pp. 697–702.

Stenner, P., Greco, M. and Motzkau, J. (2017) 'Introduction to the special issue on liminal hotspots', *Theory and Psychology*, vol. 27, no. 2, pp. 141–146

Strickland, K., Worth, A. and Kennedy, C. (2017), 'The liminal self in people with multiple sclerosis: an interpretative phenomenological exploration of being diagnosed'. *J Clin Nurs*, 26: 1714-1724. doi:[10.1111/jocn.13593](https://doi.org/10.1111/jocn.13593)

Strouse J. and Fabes R.A. (1985)' Formal versus informal sources of sex education: competing forces in the sexual socialisation of adolescence', *Adolescence* Vol. 20, No. 78, pp. 251–63.

Szakolczai, A. (2017) 'Permanent (trickster) liminality: The reasons of the heart and of the mind', *Theory and Psychology*, vol. 27, pp. 231–248.

Tanner, L. T. and Arnett, J. J. (2009) 'The Emergence of "Emerging Adulthood": The New Life Stage Between Adolescence and Young Adulthood', *Routledge Taylor & Francis Group*, Chapter 5, pp 39-45 (Accessed 12 April 2018).

Taylor, B. (2011) *The impact of assistive equipment on intimacy and sexual expression* British Journal of Occupational Therapy 74(9)bj.o.sagepub.com (Accessed March 26, 2015).

Taylor, B. (2012) 'Couples Living in Twilight: A Heideggerian Hermeneutic Study of Sexuality and Intimacy in Life-limiting Illness', PhD thesis, *Oxford Brookes University*.

Tepper, M.S. (2000) 'Sexuality and disability: the missing discourse of pleasure', *Sexuality and Disability*, 18: 4 pp 283–290.

The Times (2018) 'Obituary: Professor Stephen Hawking', 14 March [Online]. Available at <https://www.thetimes.co.uk/article/stephen-hawking> (Accessed 15 March 2018).

Thomas, A.P., Bax, M.C.O., and Smyth, D.P.L (1989) The health and social needs of young adults with physical disabilities. London Mackeith Press.

Thomas, W. I. and Znaniecki, F. (1918) 'Polish Peasant in Europe and America', *Vol. 1*, Boston MA: Gorham Press.

Thurston, C. (2009) 'The life and transitional experiences of eight young people with cystic fibrosis (CF)', Doctoral Thesis, *Anglia Ruskin University*.

Tilley, E., Walmsley, J. and Earle, S. (2012) 'The silence is roaring': sterilization, reproductive rights and women with intellectual disabilities', *Disability and Society*, vol. 27, no. 3, pp. 413–426.

Tilley, L., Walmsley, J., Earle, S. and Atkinson, D. (2012) 'International perspectives on the sterilisation of women with intellectual disabilities', in Earle, S., Komaromy, C. and Layne, L. (eds) *Understanding Reproductive Loss*, pp. 23–36, Ashgate.

Tinder (2018) [see https://tinder.com/](https://tinder.com/) (Accessed 10th March 2018)

Together for Short Lives (2013) *A Core Care Pathway for Children with Life-limiting and Life-threatening Conditions*, 3rd edn, Bristol, UK

Together for Short Lives (2015) 'Stepping Up: A Guide to Enabling a Good Transition to Adulthood for Young People with Life-limiting and Life-threatening Conditions', Bristol.

Together for Short Lives (2017) 'Guide to the Development of Children's Palliative Care Services', 4th edition, Bristol, UK.

Turner, V. (1969) 'The Ritual Process Structure and Anti-Structure', Middlesex, *Penguin*.

Turner, V. (1974) 'Dramas, Fields and Metaphors Symbolic Action in Human Society', *Ithaca, Cornell University Press*.

Trusson, D., Pilnick, A. and Roy, S. (2016) 'A new normal?: women's experience of biographical disruption and liminality following treatment for early stage breast cancer', *Social Science and Medicine*, Vol. 151, pp. 121–29.

Valentine, G. (2007) 'Theorising and researching intersectionality: a challenge for feminist geography', online at <http://doi.org/10.1111/j.1467927200587>, accessed 4 November 2016.

Van Gennep, A. ([1909]1960) 'The Rites of Passage', London, Routledge and Kegan Paul.

Volkart, E.H. (1951) 'Social behaviour and personality: contributions of W.I Thomas to theory of social research'. New York: *Social Science Research Council*.

Wade, H. A. (2002) 'Discrimination, Sexuality and People with Significant Disabilities: Issues of Access and The Right to Sexual Expression in the United States', *Disability Studies Quarterly* 22(4): 9-27, accessed 25 January 2019.

Walker, J. L. (2001) 'A qualitative study of parents' experiences of providing sex education for their children: The implications for health education', *Health Education Journal*, vol. 60, no. 2, pp. 132–146.

Walker, J. L. (2007) 'Parents and sex education—looking beyond “the birds and the bees”', *Sex Education*, vol. 4, no. 3, pp. 239–254 [Online]. DOI: [10.1080/1468181042000243330](https://doi.org/10.1080/1468181042000243330) (Accessed 12 February 2018).

Walsh, M. and Wiggins, L. (2003) 'Introduction to research: Foundations in nursing and Health Care', *Nelson Thornes, UK*.

Wascher, T. M. (2012) 'Cervical Spondylosis with Myelopathy' [Online]. Available at <https://www.spine-health.com/conditions/neck-pain/cervical-s...> (Accessed 30 January 2018).

Watts, L. (online) 'Lucy's Light'. Available at <http://www.lucy-watts.co.uk/2016/11/be-your-own-kind-of-beautiful.html> set up in 2014, (accessed 15 January 2018).

Weeks, J. (1998) 'The sexual citizen', *Theory, Culture and Society*, vol. 15, no. 3–4, pp. 35–52.

Weeks, J. (1985) 'Sexuality and its Discontents: Meanings, myths and Modern' Sexualities. London: Routledge

Weeks, J. (1986) 'Sexuality'. Chichester: Ellis Horwood/*Tavistock Publications*

Weick, K. E. (1995) 'Sensemaking in Organisations', Thousand Oaks, London, *Sage Publications*.

Williams, S. (2014) 'An Exploration of the Experiences of Fathers of Children with Disabilities Reinvention': *International Journal of Undergraduate Research*. Vol 7. Issue 2, (accessed December 12th 2018)

Wilson, B. (1991) 'The effect of drugs on male sexual function and fertility', *The Nurse Practitioner*, vol. 16, no. 9, pp. 12–17 and 21–24.

World Health Organization (WHO) (2002) 'Defining Old Age, Geneva: World Health Organization' Available at www.who.org (Accessed 13 November 2017).

World Health Organization (WHO) (2011) 'World Report on Disability', Geneva: World Health Organization [Online]. Available at www.who.org (Accessed 21 January 2018).

World Health Organization (WHO) (2015) 'Sexual Health, Human Rights and the Law', Geneva: *World Health Organization* [Online]. Available from http://apps.who.int/iris/bitstream/10665/175556/1/9789241564984_eng.pdf?ua=1 (Accessed 15 March 2018).

Yin, R. K. (2009) 'Case Study Research: Design and Methods', 4thedn, London, *Sage Publications Ltd*.

Young, B., Dixon-Woods, M., Findlay, M. & Heney, D. (2002)' Parenting in a crisis: conceptualising mothers of children with cancer'. *Social Science and Medicine* 55(10), 1835– 1847.

Zeng, Y. C., Lio, X and Loke, A.Y. (2012) 'Addressing sexuality issues of women with gynaecological cancer: Chinese nurses' attitudes and practices', *Journal of Advanced Nursing*, Vol.68 No. 2, pp. 280–92.

Appendix A: Abbreviations and definitions

In Chapters 3 to 8, I use the following abbreviations when referring to the four groups of participants:

Young adult female (*YAF*)

Young adult male (*YAM*)

Young adult – when referring to both genders (*YA*)

Parents – all were female (*MF*)

Partner (*PF* or *PM*, or collectively *Ps*)

Care practitioner – all were female (*CP*, or collectively *CPs*)

Occasionally, I refer to care practitioners as care professionals in quotations. Unless otherwise referenced, permission to reproduce or adapt the glossary has been granted by Together for Short Lives Charity and The Open University Sexuality Alliance.

Attention Deficit Hyperactivity Disorder (ADHD)

ADHD affects children and teens and may continue into adulthood. ADHD is the most commonly diagnosed mental disorder of children. Children with ADHD may be hyperactive and unable to control their impulses, or they may have trouble paying attention.

Biographical disruption

‘Chronic illness is conceptualised as a particular type of disruptive event. This disruption highlights the resources (cognitive and material) available to individuals, modes of explanation for pain and suffering, continuities and discontinuities between

professional and lay thought, and sources of variation in experience.’ (Bury, M., 1982, p. 162)

Cervical spondylotic myelopathy (CSM)

CSM refers to impaired function of the spinal cord caused by degenerative changes of the discs and facet joints in the cervical spine or neck (Wascher, 2012) and causes tingling, and temporary or intermittent paralysis.

Child

A young person aged up to their 18th birthday.

Children and young people

The term ‘children’ is used to describe babies, children and young people up to their 18th birthday.

Children’s and young people’s hospice service

Children’s hospice services provide palliative care for children and young people with life-limiting conditions and their families. Delivered by a multidisciplinary team and in partnership with other agencies, children’s hospice services take a holistic approach to care, aiming to meet the needs of both child and family – physical, emotional, social and spiritual – through a range of services. Children’s hospice services deliver this care in the home (commonly termed ‘hospice at home service’) and/or in a purpose-built building.

www.togetherforshortlives.org.uk/professionals/childrens_palliativecareessentials/definitions

Children's palliative care

Children's palliative care is an active and total approach to care, from the point of diagnosis or recognition throughout the child's life, death and beyond. It embraces physical, emotional, social and spiritual elements and focuses on enhancement of quality of life for the child/young person and support for the family. It includes the management of distressing symptoms, provision of short breaks and care through death and bereavement.

Cisgender

The same gender as acquired at birth.

Commissioner

A person with responsibility for commissioning services from service providers in the public, private or voluntary sector.

Commissioning

The process of improving outcomes and meeting the needs of the population within the local health community with the resources available.

Community services

Community services refer to a service that an individual or organisation performs within the local community. This might include community children's nurses (CCNs) who deliver nursing care and support within the local community, including visiting a patient's home. Community services may also include some of the services delivered by the local council.

Complex or continuing care

‘A bespoke package of care beyond what is available through core and universal services. It is provided to children and young people with high levels of complexity or intensity of nursing care needs’.

A CT scan

This is a Computerised Tomography scan. It is a special type of X-ray using a scanner and computer equipment to take pictures of the brain or spine in children and adults.

Depo-Provera

An injectable contraceptive, medroxyprogesterone acetate, as a form of birth control – prevents pregnancy for up to three months by stopping ovulation (the release of an egg by the ovaries) (Tilley et al., 2012).

Direct payments

Direct payments and personal budgets are offered by local authorities to enable more flexibility over how care and support is arranged and provided throughout the UK. They are delivered through a new system of Self-Directed Support in Scotland with similar but separate legislation. They are given to both people with care and support needs, and also to carers. A personal budget or direct payment will be created after an assessment by social services.

Disability or a disabled person

A person is disabled under the Equality Act 2010 if they have a physical or mental impairment that has a 'substantial' and 'long-term' negative effect on their ability to do normal daily activities. The Equality Act 2010 doesn't apply in Northern Ireland.

Disabled people and people with disabilities

These are used in primary legislation (e.g. Equality Act, 2010; Disability Discrimination Act, 1995, Northern Ireland), policy literature (e.g. Together for Short Lives, Dying Matters and Hospice UK) and by disability rights movements (e.g. The National Council for Disabled Children (NCDC)). The Equality Act, 2010, defines disability as 'a physical or mental impairment that has a substantial and long-term negative effect on a person's ability to do normal daily activities' (www.gov.uk/definition-of-disability-under-equality-act-2010).

Dopamine

The main brain chemical responsible for driving motivation
(<https://bebrainfit.com/dopamine-deficiency/>).

Dysphoria

A state of unease or dissatisfaction and may accompany anxiety, agitation and depression. It may mean that someone feels uncomfortable in and about their body
(<https://www.ncbi.nlm.nih.gov/pubmedhealth/PMHT0024720/>).

Dyspraxia

Dyspraxia is a form of developmental coordination disorder (DCD), affecting fine and/or gross motor coordination in children and adults. It may also affect speech. DCD is a lifelong condition, formally recognised by international organisations including the World Health Organisation. DCD is distinct from other motor disorders such as cerebral palsy and stroke and occurs across the range of intellectual abilities. Individuals may vary in how their difficulties present and these may change over time.

<https://dyspraxiafoundation.org.uk/aboutdyspraxia/> Accessed on February 18th, 2019)

Emerging adulthood

A distinct developmental stage spanning late adolescence to mid/late twenties, and sums up the period of ongoing transition into adulthood (Beresford, 2013).

End of life

The phase 'end of life' ends in death. Definition of its beginning is variable according to the individual child/young person and professional perspectives. In some cases, it may be the child/young person or family who first recognises its beginning. In other cases, the principal factor may be the judgement of the health/social care professional/team responsible for the care of the child/young person.

End-of-life care

End-of-life care is care that helps all those with advanced, progressive and incurable illness to live as well as possible until they die. It focuses on preparing for an anticipated death and managing the end stage of a terminal medical condition. This includes care during and around the time of death and immediately afterwards. It enables the supportive and palliative care needs of both the child/young person and

the family to be identified and met throughout the last phase of life and into bereavement. It includes management of pain and other symptoms, and the provision of psychological, social, spiritual and practical support.

Erections

Erections occur when the penis fills with blood and stretches in shape and in size. Erections can last a couple of minutes or much longer, depending on the stimulation involved. Young men typically begin getting erections during puberty as a normal part of their sexual development. Guys may also experience erections during their sleep, which can sometimes result in a 'wet dream', which occurs if one ejaculates while sleeping. www.youngmenshealthsite.org/guides/erections/

Extreme pornography

Sections 63–67 of the Criminal Justice and Immigration Act 2008 makes it an offence to possess *extreme pornographic images* that depict acts which threaten a person's life; which result in or are likely to result in serious injury to a person's anus, breasts or genitals; bestiality; or necrophilia. The Act also provides for the exclusion of classified films and sets out defences and the penalties for the offence. Section 68 and Schedule 14 of the Act are in place to ensure that the operation of the extreme pornography offence is consistent with the UK's commitments under the E-Commerce Directive (Directive 2000/31/EC) with regard to services provided by the internet industry.

Family

The term 'family' includes parents and other family members involved in the care of the young person, or other carers who are acting in the role of parents (Skills for

Health, CYP08, 2006). 'Family' includes informal carers and all those who matter to the child/young person.

Fertility

Fertility is when there is potential for reproduction. Infertility is when a couple can't get pregnant (conceive) despite having regular unprotected sex. Fertility and trying to conceive (TTC) - Live Well - NHS Choices

<https://www.nhs.uk/Livewell/Fertility/Pages/Fertilityhome.aspx> by NHS Choices - 2017 (accessed 26 April 2018)

Genetic counselling

This is the process by which individuals or relatives at risk of an inherited condition receive advice on the consequences and nature of the genetic disorder, the probability of developing or transmitting it, and the options open to them in management and family planning.

Genetic screening

This is the study of a person's DNA in order to identify genetic differences or susceptibility to particular diseases, conditions or abnormalities.

Gillick competency and Fraser guidelines

These guidelines refer to a legal case which looked specifically at whether doctors should be able to give contraceptive advice or treatment to under-16 year olds without parental consent. Since then, they have been more widely used to help assess whether a child has the maturity to make their own decisions and to understand the

implications of those decisions. www.nspcc.org.uk/preventing-abuse/child-protection-system/legal-definition-child-rights-law/gillick-competency-fraser-guidelines/ (accessed 6 October 2015).

Hegemonic

Dominant in a social context, meaning powerful (Barbour, 2014).

Hospice at home

Hospice at home is a term commonly used to describe a service which brings skilled, practical children's palliative care into the home environment. Hospice at home works in partnership with parents, families and other carers.

Hospital Episode Statistics (HES)

HES is a *data* warehouse containing details of all admissions, outpatient appointments and A&E attendances at NHS *hospitals* in England. This *data* is collected during a person's time at *hospital*. <http://content.digital.nhs.uk/hes>

ICD 10 codes

The International Classification of Diseases, Tenth Revision, Clinical Modification (ICD-10-CM) is a system used by physicians and other healthcare providers to classify and code all diagnoses, symptoms and procedures recorded in conjunction with hospital care.

Intimacy

Intimacy is more than being close or sexual intercourse. Intimacy can have different meanings for different people. The concept of intimacy involves a mutually consensual relationship where two individuals reciprocate feelings of trust, emotional and physical closeness toward each other.

<https://www.marriage.com/advice/intimacy/4-main-definitions-of-intimacy-and-what-they-mean-for-you/>

Infertility

Infertility is 'a disease of the reproductive system defined by the failure to achieve a clinical pregnancy after 12 months or more of regular unprotected sexual intercourse'.

<http://www.who.int/reproductivehealth/topics/infertility/definitions/en/>

Interdisciplinary care

This 'aims to improve the quality of life of individuals with a life-threatening condition (LTC) and life-shortening condition (LSC), seeking to reduce pain and distressing symptoms, and while attending to a variety of other psychological, social and spiritual needs that are often not adequately addressed by medical care, which emphasises cure or prolongation of life. Palliative care is patient centred and family focused, can complement curative and life- prolonging interventions from the time of diagnosis onward and can be provided in settings ranging from the home to hospital. Palliative care seeks to inform and support medical decision making by patients and families, to provide respite for carers, and to support family members in bereavement'. (Adapted from ACT, Together for Short Lives and the Royal College of Paediatrics and Child Health, 2003)

Intersectionality

This is a methodology of studying 'the relationships among multiple dimensions and modalities of social relationships and subject formations' (McCall, 2005).

Key working

Key working or care co-ordination is a service, involving two or more agencies, that provides disabled children, young people and their families with a system whereby services from different agencies are co-ordinated. It encompasses individual tailoring of services based on assessment of need, inter-agency collaboration at strategic and practice levels, and a named key worker for the child and their family. Families with disabled children should only have a key worker if they want one (Care Co-ordination Network UK, 2006).

Laparoscopy

A surgical diagnostic procedure used to examine the organs inside the abdomen. It is a low-risk, minimally invasive procedure. Only small incisions are made.

www.healthline.com (accessed 25 June 2017)

LGBTQ+

LGBT or GLBT are initials that stand for lesbian, gay, bisexual, transgender and questionable.

Life-limiting or life-shortening conditions

Life-limiting or life-shortening conditions are those for which there is no reasonable hope of cure and which will ultimately be fatal for children or young people. Some of these conditions cause progressive deterioration, rendering a child increasingly dependent on parents and carers.

Life-threatening conditions

Life-threatening conditions are those for which curative treatment may be feasible but can fail, such as children with cancer. Children in long-term remission or following successful curative treatment are not included.

Liminality

This is the ambiguity or disorientation where individuals no longer hold the status they had before the onset of their LLTC but have not yet begun the transition to the status they will hold as a result of their LLTC. Thus, according to Turner, 'betwixt and between' refers to his explanation of feeling 'neither here nor there'.

A living will

Where you can indicate whether you do or do not want interventions such as cardiac resuscitation, tube feeding or mechanical respiration.

Masturbation

This involves sexually arousing yourself by touching your genitals. Both men and women can masturbate, and you can masturbate yourself or someone else.

Masturbation is generally regarded as normal behaviour. It can help you to learn what you like or don't like sexually. Men may also use masturbation to learn how to control their orgasms, while women can find out what helps them to achieve an orgasm. Some couples masturbate together and find it a very enjoyable part of their relationship. Other people don't and that's fine. It's a personal choice.

<http://www.nhs.uk/Livewell/Goodsex/pages/masturbation.aspx>

Menstruation and the menstrual cycle

Menstruation is the vaginal bleeding that occurs in adolescent girls and women as a result of hormonal changes. It normally happens once every month. Menstruation is part of the menstrual cycle, which helps a woman's body prepare for the possibility of pregnancy each month. The parts of the body involved in the menstrual cycle include the uterus and cervix, the ovaries, fallopian tubes, the brain and pituitary gland, and the vagina. Certain body chemicals known as hormones rise and fall during the month, causing the menstrual cycle to occur.

www.healthofchildren.com/M/Menstruation.html#ixzz4q7HlyNeA

Mental disorder

The Sexual Offences Act 2003 uses the definition of mental disorder from section 1 of the Mental Health Act 1983. Section 1 of the Mental Health Act sets out that mental disorder includes any 'disorder or disability of the mind'. A person with a learning disability is not normally considered by reason of that disability to be suffering from a mental disorder unless the disability is associated with abnormally aggressive or seriously irresponsible conduct. Dependence on alcohol or drugs is not considered to be a disorder or a disability.

Metadata

Research data that describes and gives information about other data.

Meta-learning

Meta learning is described by Maudsley (1979) as "the process by which learners become aware of and increasingly in control of habits of perception, inquiry, learning, and growth that they have internalized."

Motor neurone disease (MND)

Motor Neurone Disease may affect people of any age, but most commonly occurs around 60 years and, in inherited cases, around the age of 50. The average survival from onset to death is two to four years (Rare Diseases UK, 2018). About 10% survive longer than ten years (Genetic Diseases UK, 2018). Most people die from respiratory failure. In Europe and the United States, the disease affects about two to three people per 100,000 per year (Motor Disease Association, 2018). Professor Hawking gradually became quadriplegic and required ventilation due to his advanced ALS, which affects the nerves controlling voluntary skeletal muscles but not necessarily erectile function. This is a fatal, rapidly progressing neurological disease. It attacks the nerves that control movement (motor neurones) so that muscles degenerate and no longer work, effecting walking, speaking, swallowing and breathing.

<https://brainresearchuk.org.uk/our-research/motor-neurone-disease/>.

An older person

'In most developed world countries, is usually accepted as the chronological age of 65 years. It is usually associated with the age at which one can begin to receive pension benefits, the age of which is likely to increase in future as people live and work longer. There is no general agreement on the age at which a person becomes old. The common use of a calendar age to mark the threshold of old age assumes equivalence

with biological age, yet at the same time, it is generally accepted that these two are not necessarily synonymous.'

<http://www.who.int/healthinfo/survey/ageingdefnolder/en/2013>

Outcomes

In health provision, this is the *change* in a patient's current and future health status that can be attributed to preceding health care.

Palliative care

This is the active total care of the child's body, mind and spirit, and also involves giving support to the family. It begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease. Health providers must evaluate and alleviate a child's physical, psychological and social distress. Effective palliative care requires a broad multidisciplinary approach that includes the family and makes use of available community resources; it can be successfully implemented even if resources are limited' (World Health Organization, 2002).<http://www.who.int/healthinfo/palliativecare/en/2013>

Parents

The term 'parents' has been used throughout the thesis. It is used here to mean any biological carer for a teenager or young adult, whether that is a married or unmarried couple, a single parent, guardian or foster parent. More broadly, parents or carers refers to people with parental responsibility for a child or young person. (NICE's guidelines on end-of-life care for infants, children and young people with life-limiting conditions, 2017, page 8.)

Percutaneous endoscopic gastrostomy (PEG)

This is when a tube (**PEG tube**) is passed into a patient's stomach through the abdominal wall, most commonly to provide a means of feeding when oral intake is inadequate

Polycystic ovaries

When the ovaries become enlarged and contain many fluid-filled sacs (follicles) which surround the eggs. www.nhs.uk/conditions/polycystic-ovarian-syndrome/Pages/Introduction.aspx

Power of attorney

A power of attorney directive names someone who you trust to act as your agent if you are unable to speak for yourself. If you want to choose one person to speak for you on health care matters, and someone else to make financial decisions, you can do separate financial and health care powers of attorney.

Precocious puberty

The onset of signs of puberty before age seven or eight in girls, and age nine in boys. In girls, this may include any of the following before seven or eight years: breast development, pubic or underarm hair development, a rapid growth in height, start of menstruation, acne and a mature body odour. In boys, signs before nine years of age include: enlargement of the penis or testicles, pubic, underarm or facial hair development, rapid growth in height, deepening voice, acne and a mature body odour.

Primary healthcare team

Comprises the general practitioner, practice nurse and community staff (such as community children's nurses or physiotherapists) who work with the practice staff.

Precocious (Early Puberty) and Delayed Puberty

Early puberty, also called precocious puberty, is when girls have signs of puberty before 8 years of age and boys have signs of puberty before 9 years of age. Some girls and boys may develop certain signs of puberty at a young age, but not others. For example, girls may start periods before the age of 8 but have no breast development. By contrast, delayed puberty is when boys have no signs of testicular development by 14 years of age and girls have not started to develop breasts by 13 years of age, or they have developed breasts but their periods have not started by 15 years. See: <https://www.nhs.uk/conditions/early-or-delayed-puberty/>.

Puberty

The period during which normal/normative adolescents usually reach sexual maturity and become capable of reproduction.

Reproduction

This is a fundamental feature of all known life; each individual organism exists as the result of reproduction. There are two forms of reproduction: asexual and sexual. In asexual reproduction, an organism can reproduce without the involvement of another organism whereas sexual reproduction requires another individual/being.

Sex

Refers to the biological characteristics that define humans as female or male. While these sets of biological characteristics are not mutually exclusive, as there are individuals who possess both, they tend to differentiate humans as males and females. In general use in many languages, the term sex is often used to mean 'sexual activity', but for technical purposes in the context of sexuality and sexual health discussions, the above definition is preferred. This may also include sexual activity, including specifically sexual intercourse.

www.who.int/reproductivehealth/topics/sexual_health/sh_definitions/e/ (accessed 6 October 2017).

Sexual Consent

The age of sexual consent refers to the minimum age at which a person is considered to have the legal capacity to consent to sexual intercourse, whereas the legal age of marriage is the minimum age at which a person is deemed able to consent to marry.

Sexual Identity

One part of the umbrella concept of sexual orientation. It does not reflect sexual orientation or sexual behaviour- these are separate concepts which the OFFICE FOR (National Statistics UK (ONS) currently does not measure. (ONS, 2015, p.2)

Sexual health

'...a state of physical, emotional, mental and social wellbeing in relation to sexuality; it is not merely the absence of disease, dysfunction or infirmity. Sexual health requires a positive and respectful approach to sexuality and sexual relationships, as well as the

possibility of having pleasurable and safe sexual experiences, free of coercion, discrimination and violence. For sexual health to be attained and maintained, the sexual rights of all persons must be respected, protected and fulfilled.’ (WHO, 2006a)

Sexuality

A central aspect of being human throughout life encompasses sex, gender identities and roles, sexual orientation, eroticism, pleasure, intimacy and reproduction. Sexuality is experienced and expressed in thoughts, fantasies, desires, beliefs, attitudes, values, behaviours, practices, roles and relationships. While sexuality can include all these dimensions, not all of them are always experienced or expressed. Sexuality is influenced by the interaction of biological, psychological, social, economic, political, cultural, legal, historical, religious and spiritual factors.” (WHO, 2006a)

Sexual rights

There is a growing consensus that sexual health cannot be achieved and maintained without respect for, and protection of, certain human rights. The working definition of sexual rights given below is a contribution to the continuing dialogue on human rights related to sexual health.

‘The fulfilment of sexual health is tied to the extent to which human rights are respected, protected and fulfilled. Sexual rights embrace certain human rights that are already recognized in international and regional human rights documents and other consensus documents and in national laws.

Rights critical to the realization of sexual health include:

- the rights to equality and non-discrimination

- the right to be free from torture or to cruel, inhumane or degrading treatment or punishment
- the right to privacy
- the rights to the highest attainable standard of health (including sexual health) and social security
- the right to marry and to found a family and enter into marriage with the free and full consent of the intending spouses, and to equality in and at the dissolution of marriage
- the right to decide the number and spacing of one's children
- the rights to information, as well as education
- the rights to freedom of opinion and expression, and
- the right to an effective remedy for violations of fundamental rights.

The responsible exercise of human rights requires that all persons respect the rights of others. The application of existing human rights to sexuality and sexual health constitute sexual rights. Sexual rights protect all people's rights to fulfil and express their sexuality and enjoy sexual health, with due regard for the rights of others and within a framework of protection against discrimination.' (WHO, 2006a, updated 2010)

Specialist palliative care

Specialist palliative care is defined as the total care of patients with progressive, far advanced disease and limited prognosis and their families, by a multi-professional team who have undergone recognised specialist palliative care training. It provides physical, psychological, social and spiritual support, and will involve practitioners with a broad mix of skills (NICE, 2004). These services are provided by statutory and voluntary organisations and cover community, hospice and hospital inpatient settings.

Supportive care

Supportive care is an 'umbrella' term for all services, both generalist and specialist, that may be required to support people who have a life-threatening illness. It is not a response to a particular disease or its stage but is based on an assumption that people have needs for supportive care from the time that the possibility of a life-threatening condition is raised. LEA

Technology-dependent children

Children who need both a medical device to compensate for the loss of a vital bodily function and substantial and ongoing nursing care to avert death or further disability.

What isn't counted as a disability?

Some conditions aren't covered by the disability definition. These include addiction to non-prescribed drugs or alcohol.

Young adult

This term describes a person between their 19th and 40th birthdays (The Open University Sexuality Alliance, 2016, p. 6)

Young person

This term describes a person from their 13th to their 19th birthday.

Appendix B: An overview of current UK Law (at the time of writing)

Adapted and updated with permission from The Open University Sexuality Alliance

Young people with LLTCs should have the same right to relationships, fun and sexual expression as anyone else as long as no harm will be caused (De Than, 2014).

‘wrapping [disabled people] up in cotton wool... The fact is that all life involves risk... physical health and safety can sometimes be bought at too high a price in happiness and emotional welfare.’ (Munby, 2007)

Important statutes in England and Wales:

- The Care Act 2014
- The Children Act 1989
- The Children and Families Act 2014
- The Equality Act 2010
- The Equality Act 2010 (Specific Duties) Regulations 2011
- The Human Rights Act 1998
- The Mental Capacity Act 2005
- The Sexual Offences Act 1956, 1967, 2003, 2006, 2009

Important statutes in Scotland:

- The Adults with Incapacity (Scotland) Act 2000
- The Age of Legal Capacity (Scotland) Act 1991
- The Equality Act 2010

- The Prohibition of Female Genital Mutilation (Scotland) Act 2005
- The Protection from Abuse (Scotland) Act 2001
- The Protection of Children (Scotland) Act 2003
- The Protection of Children and Prevention of Sexual Offences (Scotland) Act 2005
- The Sexual Offences (Scotland) Act 2009

Important statutes in Northern Ireland:

- The Children (Northern Ireland) Order 1995
- The Disability Discrimination Acts 1995 and 2005
- The Female Genital Mutilation (England, Wales and Northern Ireland) Order 2003
- The Human Rights Act 1998
- The Safeguarding Vulnerable Groups (Northern Ireland) Order 2007
- The Sexual Offences (Northern Ireland) Order 2008
- Female Genital Mutilation (England, Wales and Northern Ireland) 2003

International Statutes

- The European Convention on Human Rights
- The United Nations Convention on the Rights of Persons with Disabilities

The Chatham House Rule

When a meeting, or part thereof, is held under the *Chatham House Rule*, participants are free to use the information received, but neither the identity nor the affiliation of the speaker(s), nor that of any other participant, may be revealed. See

www.chathamhouse.org/about/chatham-house-rule.

The European Convention on Human Rights

The most relevant human rights law in the UK is the European Convention on Human Rights (ECHR) (see <https://rightsinfo.org/the-rights-in-the-european-convention>), particularly Articles 8, 10 and 14. Currently, under Article 8 of that Convention and incorporated into the Human Rights Act 1998 in England and Wales, *everyone* has the right to respect for their privacy, family life, home and correspondence. The ECHR indicates that Article 8 protects sexual autonomy, confidentiality, dignity, forming and maintaining personal relationships and allowing them to develop normally (*Pretty v UK* 2002). So, often staff caring for young people fear reprisal and are frightened of the law, fearing that they may risk committing an offence or colluding in a crime. Human Rights may only be limited by the State if it needs to do so to achieve an aim, such as preventing crime or upholding the rights of others. So there should be an equal right for all adults to have consensual sexual activity in private, alone or consensually with others, to have a relationship of their choice, be intimate and have a child, if they so wish (De Than, 2014).

The Equality Act 2010

The Equality Act 2010 imposes a duty on public authorities to promote equality for disabled people by treating them more favourably and, where necessary, making reasonable adjustments for people with disabilities. This could mean, for example, arguing that young people can access information, education and equipment if required at various stages of their life. A key issue in the UK is that different rules and laws are based on age, mental capacity, the nature of the disability, and whether a disabled person lives at home or in residential care. This often makes issues about sexual expression so complicated that many people don't understand what they can and cannot do.

Sexual Offences Act 2003 (SOA)

The SOA 2003 criminalises underage teenage sexual behaviour, and those who assist it, even when they act in the best interests of that teenager. Young people under 16 years cannot consent to sex or sexual touching or being touched sexually. It is also a crime for people in a position of trust, such as care staff and teachers, to have sexual contact with a child under 18 years (De Than, 2014).

Currently, a child whose disabilities hinder their sexual expression or communication (for example, being unable to masturbate) can only be supported in ways which do not involve touching the child in any way which may be interpreted as sexual. Here the criminal law conflicts with the human rights of the young person. Naturally there is a delicate balance between protecting a young person from harm and respecting their human rights, particularly their right to sexual expression.

When a young person has a LLTC which may affect communicating consent, such as hearing and speech difficulties, the SOA 2003 presumes that person does not consent to sexual activity and anyone who has sex with that young person cannot guarantee that there was consent. This would then be presumed to be a sex crime. Campaigners argue that this is an issue in need of legal reform.

The SOA 2003 potentially criminalises people in long-term relationships when one person develops or suffers brain injury, has multiple disabilities or develops a mental illness. There is case law where judges have decided that a disabled person lacked capacity to make informed decisions about sexual expression and prevented that person from continuing their relationship with another person. The SOA, Sections 30–33, creates offences against a person with a ‘mental disorder’. This can be particularly relevant where care professionals are supporting clients aged over 18.

Many staff are understandably concerned that sexual activity *might* occur during their work shift. Unless the practitioner was actively encouraging sexual activity to take place, arguably the practitioner would not be committing an offence. However, there is a possibility of the practitioner or organisation being sued for compensation if they knew at the time that *sexual abuse* was taking place and did nothing to prevent this, but not if consensual sexual activity was thought to be taking place (De Than, 2014).

The Mental Capacity Act (MCA) 2005

The premise of this Act is that everyone is presumed to have mental capacity to make decisions about their own lives, unless and until it is proved that they lack capacity. The Act recognises that everyone has the right to make unwise decisions and in doing so, does not necessarily indicate that a person lacks capacity. The Act does not and cannot apply to sexual matters as no one can consent to sex on behalf of another person, even though sometimes organisations believe they may be able to intervene.

The Court has stated that the basic requirements for capacity to consent to sex are as follows (*Re AB 2011 Court of Protection, EWHC 101*):

‘A person has capacity to consent to sex if they understand on a simple level:

- the mechanics of the act
- that there are health risks involved, particularly the acquisition of sexually transmitted and transmissible infections and
- (if relevant to them) that sex between a man and a woman may result in the woman becoming pregnant.’

If there is anything that can enable a young person to have that basic understanding, then it should be facilitated. It is not lawful to prevent an adult from having

consensual sexual expression in private, alone or accompanied, unless they have already been found to lack capacity to make decisions about sex.

General legal principles in the area of sexuality

Touching clients

Generally, clients should not be touched without their permission; doing so may be judged as an assault. There may be exceptions when it may not be possible to obtain consent, such as in a medical emergency. Qualified health professionals, such as doctors, dentists, nurses and physiotherapists may touch their patients within these boundaries but not in a sexual way. Sometimes the law implies consent, such as touching someone to gain their attention, or maybe to reassure or support them if the young person is distressed. Care staff who breach consent could be convicted of one or more criminal offences.

Sexual offences and care professionals

The types of potentially criminal behaviour found in the Sexual Offences Act that care professionals might be concerned about are:

1. 'causing' or 'inciting' unlawful sexual behaviour, or
2. 'aiding, abetting or counselling' that behaviour. It is important to consider what is meant by each type of offence.

1. 'Causing or inciting'

A care professional would commit a criminal offence by *intentionally* causing or inciting a young person under 16 to engage in sexual activity (see Section 10 of the

SOA). Section 31 of the SOA has a similar effect in respect of a person with a 'mental disorder' impeding choice. However, it is important to understand what 'causing' and 'inciting' mean. A care professional would only 'cause' an act if controlling or influencing the person in their care to do it. It is not enough that the care professional assisted the person in their care. Similarly, to 'incite' is to urge or encourage and not to merely assist at the request of the person in care.

2. 'Aiding, abetting or counselling'

The criminal law always recognises that it is an offence to assist another to commit an offence. However, care professionals may benefit from a number of statutory exceptions in the SOA in respect of liability for aiding, abetting or counselling the commission of sexual offences against children. These apply where the aider or abettor does not act for the purposes of sexual gratification or causing or encouraging the activity, but to protect the child from sexually transmitted infection, to preserve the child's physical safety, to prevent the child from becoming pregnant or to promote the child's emotional wellbeing by the giving of advice.

Clients with 'mental disorders'

Additionally, care professionals are subject to a specific regime in respect of those labelled by the law as having a 'mental disorder' in their care, whether that mental disorder has the effect in practice of restricting choice. The intentional sexual touching of a person with a mental disorder is an offence (Section 38 of the SOA), as is causing or inciting sexual behaviour (Section 39 of the SOA), as well as certain other matters.

Case law

- CH v A Metropolitan Council [2017] EWCOP 12

- Munby, RE MM, 2007; EWHC, 2003
- Re AB 2011 Court of Protection, EWHC 101
- Pretty v UK [2002]

Gillick v West Norfolk and Wisbech AHA [1985] UKHL

It must be noted that sexuality is a complex area of law. While these general principles apply, their application will vary depending on the specifics of a situation. Care practitioners are encouraged to seek legal advice to clarify how the law will apply in a specific situation.

Appendix C: Literature review Strategy

<i>Search engine</i>	<i>Search terms</i>	<i>Literature</i>
PubMed, the Association of Public Health Observatories, Public Health England, National Health Service Information Centre and the Department of Health, Government, UK; Charity databases and websites, international journals of sociology, other social sciences, legal, medicine and nursing, including palliative care, sexuality and disability, and other policy and grey literature, including blogs.	'life limited', 'life threatened', 'life limiting', 'life shortened conditions or illnesses in young people and adults', 'palliative care'	Clinical, Statistical, Epidemiology, Policy : government and third sector publications / websites
PubMed, the Association of Public Health Observatories	'prevalence in young people and adults with life limiting or life threatening conditions', 'aetiology', 'epidemiology'	
One Stop Open University Database, the British Library, SCONUL Access, Google Scholar, international journals of sociology, other social sciences, legal, medicine and nursing, including palliative care, sexuality and disability, and other policy and grey literature, including blogs.	'sexuality', 'sex', 'relationships', 'sexuality and disability', 'disability and reproduction', 'reproductive choices and loss', 'attitudes and choices', 'genetic counselling', 'taboo', 'sex therapy'	
international journals of sociology, other social sciences, legal, lawtel and legal citators, medicine and nursing, including palliative care, sexuality and disability journals, reproductive health journals, policy and grey literature, including blogs.	'sexuality and end-of-life care', 'sexuality and the law', 'protection from harm', 'vulnerable adults', 'life course approach', theory and perspective', 'uncertainty', 'liminality', 'biographical disruption', 'transition theories'	

Appendix D: Research project ethics approval



The Open University

From Dr Duncan Banks
Chair, The Open University Human Research Ethics Committee
Email duncan.banks@open.ac.uk
Extension 59198

To Maddie Blackburn, Health and Social Care

Subject *"The sexuality, relationships and reproductive choices of people with non-malignant life-limited conditions in the UK."*

Ref HREC/2013/1491/Blackburn/2

Red form

Submitted 8 November 2013

Date 10 November 2013

Memorandum

This memorandum is to confirm that the research protocol for the above-named research project, as submitted for ethics review, **has been given a favourable opinion** by the Open University Human Research Ethics Committee.

Please make sure that any question(s) relating to your application and approval are sent to Research-REC-Review@open.ac.uk quoting the HREC reference number above. We will endeavour to respond as quickly as possible so that your research is not delayed in any way.

At the conclusion of your project, by the date that you stated in your application, the Committee would like to receive a summary report on the progress of this project, any ethical issues that have arisen and how they have been dealt with.

Regards,

Dr Duncan Banks
Chair OU HREC

The Open University is incorporated by Royal Charter (number RC 000391), an exempt charity in England & Wales and a charity registered in Scotland (number SC 038302)

HREC_2013-#1491-Blackburn-2-approval

Appendix E: Young adults' information sheet



Sexuality, relationships and reproductive choices of young adults with life-limiting and life-threatening conditions

(Participant information for young people and adults with life-limiting and/or life-threatening conditions (age 16+))

Maddie Blackburn, PhD student
The Open University, Walton Hall
Milton Keynes, MK7 6AA
Email: maddie.blackburn@open.ac.uk www.open.ac.uk

Dear xxx

What is my research about?

Hello. My name is Maddie Blackburn. I am a doctoral research student at The Open University in Milton Keynes. I am carrying out a research study about the sexuality, relationship and reproductive choices and experiences of young adults over 16 years with conditions such as cystic fibrosis, Duchenne muscular dystrophy, spinal muscular atrophy or cancer. I am writing to ask if you would be interested in contributing to this research please. Recently, people with these conditions, as well as parents, carers and care staff, have been seeking information about relationships and sex and how and where to obtain the best advice on these matters. We hope that this research will helpfully address what information and advice would be useful about sexuality and relationships for people who have such conditions. If you agree to take part, I would like to meet with you and talk to you about the following:

- Your views about relationships, sex and reproduction
- Whether you think you need any advice or training on these matters
- If you feel it would be helpful for your parents, carers or care practitioners to know more about sex and relationships for people with conditions like yours.

What does the research involve?

A one-to-one interview with you which will probably last between one and two hours, at a mutually agreed time and place, discussing these issues.

A second interview with you may be requested at a later date following all of the other interviews with different people in this research. The Hospice has kindly offered to provide a room for the interview which will be with me. You can bring someone to the interview with you if you wish. The interview will be recorded and transcribed by me and any report of it will be carefully anonymised, in other words, any names removed. Care will be taken to ensure that additional individual identifying details are not included in any public domain media.

As part of my research, I shall also be separately interviewing other people. In addition, I shall be interviewing parents and carers who help support people with similar conditions, although these will not necessarily be your own parents or carers. I shall also be interviewing care staff at Douglas House.

Will the information I collect remain private?

Each person who takes part in this study will be asked to sign a consent form (see Appendix H). I will not use your name in any notes I make or any information I record or store for writing up my research. Your views and contributions will be treated

confidentially. The information will always be securely stored, and the research findings are published.

Can you leave the research?

Yes. You are free to leave the research without reason, at any time, if you no longer wish to take part. You may also ask me to exclude or edit some or part of the data collected. If you choose to leave the research before I begin analysing the information, all of the information you have given me will be destroyed, unless you consent to the information being used in this study. Once I start analysing the information I have obtained from you, I shall ask your permission to continue using the information for this research. Also, if there are any questions that you don't particularly want to discuss during the interview, you don't have to. You can ask me to move on to another question or discussion point.

Do you have to take part in both interviews?

No. If you only want to take part in one interview, that is fine. However, I shall ask your permission to use the information from the first interview for the purpose of the research and will separately invite you to a second interview.

Will there be any risks associated with you taking part?

I do not anticipate that there will be risks associated with this research. I shall be happy to discuss anything with you immediately after the interview or on another day, if that suits you better. I can provide you with my contact details and names and

details of organisations should you require any further information or support about some of the topics we discuss.

How will the research be used in future?

Results of this research may be published in future, but no details of individuals or institutions will be identifiable.

If you want to contribute

I do hope that you will take part in this research. Please do feel free to contact me if you would like to discuss any aspect of this research or require any further information before agreeing to take part. I may be contacted by email, details of which are supplied at the top of this letter.

If you want to talk to someone else about this research from The Open University, you can contact the Associate Dean (Research and Enterprise): **Dr Sarah Earle, at the Faculty of Health and Social Care, The Open University, Walton Hall, Milton Keynes, MK7 6AA.**

If you do agree to take part in this research, please could you let me know on what date and time it would be convenient for me to meet with you please either via email, phone or via The House Manager at Douglas House.

Many thanks indeed for considering this request.

Yours sincerely,

Maddie Blackburn

PhD Student

Research supervisors: Dr Sarah Earle: sarah.earle@open.ac.uk and Dr Carol

Komaromy: carol.komaromy@open.ac.uk

Appendix F: Parents' and partners' information sheet

Sexuality, relationships and reproductive choices of young adults with life-limiting and life-threatening conditions

(Participant information for Parents / Partners)

Maddie Blackburn, PhD student
The Open University, Walton Hall
Milton Keynes, MK7 6AA
Email: maddie.blackburn@open.ac.uk www.open.ac.uk

Dear xxx

What is my research about?

My name is Maddie Blackburn. I am a research student at The Open University in Milton Keynes. I am carrying out a research study about the sexuality, relationship and reproductive experiences and choices of young adults over 16 years with life-limiting conditions, such as cystic fibrosis, Duchenne muscular dystrophy and spinal muscular atrophy. I am writing to ask if you would be interested in contributing to this research please. Recently several young people with life-limiting conditions, as well as care staff and other parents, have been seeking information about relationships and sex and how and where to obtain the best advice on these matters for young adults with life-limiting conditions. We hope that this research will helpfully address what information and advice would be useful. If you agree to take part, I would like to meet with you and talk to you at a mutually agreed date, time and place about the following:

- Your views about relationships, sex and reproduction in people with life-limiting conditions.

- Whether you think either you or your son/daughter would benefit from advice or training on these matters.
- If you feel it would be helpful for other carers or staff to know more about sex, reproduction and relationships for people with life-limiting conditions.

What does it involve?

A one-to-one interview with you which will probably last between one and two hours, at a mutually agreed time and place, discussing these issues. You are welcome to bring someone to the interview with you if this would be helpful. The interview will be recorded and transcribed by me and any report of it will be carefully anonymised. Care will be taken to ensure that additional individual identifying details are not included in any public domain media.

As part of my research, I shall be separately interviewing young adults with life-limiting conditions, care staff and other parents and carers who support these young people.

Will the information I collect remain private?

Each person who takes part in this study will be asked to sign a consent form (see Appendix H). I will not use your name in any notes I make or any information I record or store for writing up my research. Your views and contributions will be treated confidentially. The information will be securely stored at all times and deleted after the research is completed and the research findings are published. All of the information I collect will be anonymised (nameless) and will be safely stored on

password-protected, encrypted computers and Iron Key memory sticks. Any hard copies of information will be safely stored in locked cabinets.

Can you leave the research?

Yes. You are free to leave the research without reason, if you feel you no longer wish to take part. You may also ask me to exclude or edit some of the data collected. If you choose to leave the research before I begin analysing the information, all of the information you have given me will be destroyed, unless you consent to the information being used in this study. Once I start analysing the information, I shall seek your permission to continue using that information for the research. Also, if there are any questions that you do not want to discuss during the interview, you don't have to. You can ask me to move on to another question or discussion point.

How many interviews will I need to attend?

You will be invited to participate in one interview only.

Does this research conform to ethical guidelines?

The ethics for my project has received favourable opinion by The Open University's Human Research and Douglas House Ethics Committees. This means that the topics and methods of my research have been reviewed by the Ethics Committee and judged to be in compliance with The Open University's research ethics principles.

Will there be any risks associated with you taking part?

I do not anticipate that there will be risks associated with this research. I have received a clear enhanced disclosure from the Disclosure and Barring Service (DBS),

formerly known as a Criminal Records Bureau. I shall also be happy to discuss anything with you immediately after the interview or on another day if that suits you better. I can provide you with my contact details and names and details of organisations and services should you require any further information about some of the topics we discuss.

How will the research be used in future?

Results of this research may be published in future in both professional and charity journals, such as Together for Short Lives' *Family Care Matters*, but no details of individuals or institutions will be identifiable.

If you want to contribute

I do hope that you will wish to take part in this research. Please do not hesitate to contact me if you would like to discuss any aspect of this research or require any further information before agreeing to take part. I may be contacted by email, details of which are supplied at the top of this letter. If you want to talk to someone else about this project, you can contact the Associate Dean (Research and Enterprise): **Dr Sarah Earle at the Faculty of Health and Social Care, The Open University, Walton Hall, Milton Keynes, MK7 6AA.**

If you do agree to take part in this research, please could you let me know on what date and time it would be convenient for me to meet with you either via email, phone or via the House Manager at Douglas House Hospice. I shall require you to sign the consent form below in advance of the interview.

Many thanks indeed for considering this request.

Maddie Blackburn

PhD Student

Research supervisors: Dr Sarah Earle: sarah.earle@open.ac.uk and Dr Carol Komaromy: carol.komaromy@open.ac.uk

Appendix G: Care practitioners' information sheet



Sexuality, relationships and reproductive choices of young adults with life-limiting and life-threatening conditions

(Participant information for Care Practitioners)

Maddie Blackburn, PhD student
The Open University, Walton Hall
Milton Keynes, MK7 6AA
Email: maddie.blackburn@open.ac.uk www.open.ac.uk

Dear xxx

What is my research about?

My name is Maddie Blackburn. I am a research student at The Open University in Milton Keynes. I am carrying out a research study about the sexuality, relationship and reproductive experiences of young adults over 16 years with life-limiting or life-threatening conditions, such as cystic fibrosis, Duchenne muscular dystrophy, spinal muscular atrophy or cancer. I am writing to ask if you would be interested in contributing to this research please. There is limited research addressing the sexuality of young adults over 16 years with life-limiting conditions (LLTCs). This is because, until recently, children life-limiting or life-threatening conditions were not expected to reach adulthood but now are, so relationships, sexuality and reproduction were not often discussed with families or care staff. Recently several young people with life-limiting conditions, as well as care staff and other parents, have been seeking information about relationships and sex, and how and where to obtain the best advice

on these matters. We hope that this research will helpfully address what information and advice would be useful for young people with LLTCs. If you agree to take part, I would like to meet with you at a mutually convenient place, date and time to talk to you about the following:

- Your views about relationships, sex and reproduction for people with life-limiting conditions
- Whether you think care staff would benefit from advice or training on these matters
- If you feel it would be helpful for parents and other carers to know more about sex, reproduction and relationships for people with life-limiting conditions.

What does it involve?

A one-to-one interview with you which will probably last between one and two hours, at a mutually agreed time and place, discussing these issues. The interview will be with me. You are welcome to bring someone to the interview with you if this would be helpful. The interview will be recorded and transcribed by me and any report of it will be carefully anonymised. Care will be taken to ensure that additional individual identifying details are not included in any public domain media.

As part of my research, I shall be separately interviewing people with life-limiting conditions, care staff and other parents and carers.

Will the information I collect remain private?

Each person who takes part in this study will be asked to sign a consent form (see Appendix H). I will not use your name in any notes I make or any information I record or store for writing up my research. Your views and contributions will be treated

confidentially. The information will be securely stored at all times and deleted after the research is completed and the research findings are published. All of the information I collect will be anonymised and will be safely stored on password-protected, encrypted computers and Iron Key memory sticks. Any hard copies of information will be safely stored in locked cabinets.

Can you leave the research?

Yes. You are free to leave the research without reason, if you feel you no longer wish to take part. You may also ask me to exclude or edit some of the data collected. If you choose to leave the research before I begin analysing the information, all of the information you have given me will be destroyed, unless you consent to the information being used in this study. Once I start analysing the information, I shall seek your permission to continue using that information for the research. Also, if there are any questions that you do not want to discuss during the interview, you don't have to. You can ask me to move on to another question or discussion point.

How many interviews will I need to attend?

You will be invited to participate in one interview only.

Does this research conform to ethical guidelines?

The ethics for my project has been approved by The Open University's Human Research Ethics Committee and has been given a favourable opinion. This means that the topics and methods of my research have been reviewed by the Ethics Committee and judged to be in compliance with The Open University's research ethics principles. The work is also being supervised by Drs Earle and Komaromy, whose details are included at the end of this letter.

Will there be any risks associated with you taking part?

I do not anticipate that there will be risks associated with this research. I have received a clear, enhanced disclosure from the Disclosure and Barring Service (DBS), formerly known as a Criminal Records Bureau. I shall also be happy to discuss anything with you immediately after the interview or on another day if that suits you better. I can provide you with my contact details and names and details of organisations and services should you require any further information about some of the topics we discuss.

How will the research be used in future?

Results of this research may be published in future in both professional and charity journals, such as Together for Short Lives' *'Family Care Matters'*, but no details of individuals or institutions will be identifiable.

If you want to contribute

I do hope that you will wish to take part in this research at Rainbows Hospice. Please do not hesitate to contact me if you would like to discuss any aspect of this research or require any further information before agreeing to participate. I may be contacted by email, details of which are supplied at the top of this letter. If you want to talk to someone else about this project, you can contact the Associate Dean (Research and Enterprise): **Dr Sarah Earle at the Faculty of Health and Social Care, The Open University, Walton Hall, Milton Keynes, MK7 6AA.**

If you do agree to take part in this research, please could you let me know on what date and time it would be convenient for me to meet with you either via email, phone

or via the Director of Care at Rainbows Hospice. I shall require you to sign the consent form in Appendix H in advance of the interview.

Many thanks indeed for considering this request.

Maddie Blackburn

PhD Student

Research supervisors: Dr Sarah Earle: sarah.earle@open.ac.uk and Dr Carol Komaromy: carol.komaromy@open.ac.uk

Appendix H: Consent forms



Sexuality, relationships and reproductive choices in young adults with life-limiting and/or life-threatening conditions.

Consent form one

Parents/Carers taking part in face-to-face interviews

Name of Project Researcher: Maddie Blackburn

Please initial box

I confirm that I have read and understand the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.

☐

I understand that my participation is voluntary and that I can withdraw at any time without giving any reason.

☐

I understand that the information collected in this study will be confidential, anonymous and only shared with Maddie Blackburn's Supervisors: Drs Sarah Earle and Carol Komaromy.

☐

I am happy to take part in an interview with the researcher, Maddie Blackburn.

☐

Name of participant	Date	Signature
---------------------	------	-----------

Name of person taking consent	Date	Signature
-------------------------------	------	-----------

Consent form two



(Young adults taking part in face-to-face interviews who can provide written consent)

(If the young person is unable to give written consent, the contents of the study will be carefully explained to the young adults and a file note made as to whether the young person wishes to take part in the study or not.)

Name of Project Researcher: Maddie Blackburn

Please initial box

I confirm that I have read and understand the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.

☐

I understand that my participation is voluntary and that I can withdraw at any time without giving any reason.

☐

I understand that the information collected in this study will be confidential, anonymous and only shared with Maddie Blackburn's Supervisors: Drs Sarah Earle and Carol Komaromy.

☐

I am happy to take part in an interview with the researcher, Maddie Blackburn.

☐

Name of participant

Date

Signature

Name of person taking
consent

Date

Signature

Consent form three



Care staff taking part in face-to-face interviews

Name of Project Researcher

Name of Project Researcher: Maddie Blackburn

Please initial box

I confirm that I have read and understand the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.

☐

I understand that my participation is voluntary and that I can withdraw at any time without giving any reason.

☐

I understand that the information collected in this study will be confidential, anonymous and only shared with Maddie Blackburn's Supervisors: Drs Sarah Earle and Carol Komaromy.

☐

I am happy to take part in an interview with the researcher, Maddie Blackburn.

☐

Name of participant

Date

Signature

Name of person taking
consent

Date

Signature

Appendix I: Life-limiting and/or life-threatening conditions relevant to this research

Adrenoleukodystrophy (ALD)

This is an X-linked recessive genetic life-limiting condition caused by an abnormality in a gene on the X chromosome. The prevalence is approximately 1/20,000–1/50,000 births and most affected are boys. Approximately half of all females who carry the abnormal ABCD1 gene will develop some symptoms of ALD. The condition occurs in all ethnic groups. This condition affects the white matter of the nervous system and the adrenal cortex. Some affected individuals have adrenal insufficiency, which means that reduced amounts of certain hormones such as adrenaline and cortisol are produced, leading to abnormalities in blood pressure, heart rate, sexual development and reproduction. Some of those affected experience serious neurological problems that can influence mental function and lead to disability and reduced life span. This condition has been categorised into six types based on symptoms and age of onset: childhood cerebral ALD, adolescent cerebral ALD, adrenomyeloneuropathy, adult cerebral ALD, adrenal insufficiency only and ALD that occurs in females. Childhood ALD usually begins between four and eight years of age and symptoms include attention deficit disorder, progressive loss of intellectual function, and vision, hearing and motor deterioration. Adolescent cerebral ALD begins between 11 and 21 years of age and the symptoms are like the childhood cerebral type, but the disease progresses more slowly.

<https://rarediseases.org/rare-diseases/adrenoleukodystrophy/>

Acute myeloid leukaemia

Acute myeloid leukaemia (AML) is a blood cancer that starts from young white blood cells in the bone marrow. It occurs in both adults and children. Chemotherapy is the main treatment, although a bone marrow or stem cell transplant is sometimes suggested.

Benign and malignant tumours in children and young people

Brain and spinal cord cancers are diagnosed in one in four children with cancer.

Astrocytomas are the most common type of glioma in both adults and children. They develop from cells called astrocytes in the brain that support the nerve cells (neurones). Focal astrocytomas are often diagnosed in children and young people, and are not common in adults. Other astrocytomas are called diffuse. These do not have a clear boundary between the tumour and normal brain tissue.

<http://www.cancerresearchuk.org/about-cancer/childrens-cancer/about>

Childhood cancers

Child and adolescent cancer is much less common than adult cancer. In the UK, around 1,800 children (aged 0–14 years) are diagnosed with cancer each year. This number includes non-cancerous (benign) brain tumours. Children develop different types of cancers from adults but they often receive similar treatments. The most common are acute leukaemias, diagnosed in one in three children with cancer.

Cystic fibrosis

Cystic fibrosis (CF) is one of the most common life-limiting, life-shortening, chronic fatal genetic diseases affecting around 10,000 children and young adults in the UK. It affects the lungs and digestive system, making it difficult to breathe and digest food. Complications increase with age, requiring ever increasing levels of care, treatment and support. The average life expectancy is in the late 30s. There is no cure. It is an inherited disease caused by a faulty gene and occurs in both males and females. The gene controls the movement of salt and water in and out of cells, so the lungs and digestive system become clogged with mucus, making it hard to breathe and digest food. In 1964, a child born with CF was unlikely to live beyond five years.

www.cysticfibrosis.org.uk

Duchenne muscular dystrophy

Duchenne muscular dystrophy (DMD) is a progressive, genetic, neuromuscular condition caused by the lack of a protein in a gene called dystrophy, occurring in males in about 1 in every 3,500 UK births. It's a life-limiting condition for which there is at present no known cure. DMD causes progressive muscle weakness over a number of years but the men usually have mental capacity unless there are accompanying learning difficulties.

By the age of 13, boys with DMD generally lose the ability to walk independently and by the mid-teens, there are other complications: for example, curvature of the spine, respiratory difficulties and cardiac failure. The mean age of death without specialised treatment is extending, but during the last few years there have been significant improvements in the ways the condition is treated.

About 100 males with DMD are born in the United Kingdom each year and some men are living until their third or fourth decade (Abbott, 2012c). At present, average life expectancy for people with DMD is around 27 years (Eagle et al., 2007). The mean age of death is continuing to rise as more effective medical interventions impact on the current generation of men who are living well into adulthood (Abbott et al., 2015). There are approximately 2,500 known males living with this condition in the UK at any one time and an increasing number living into adulthood. Sometimes more than one child is affected in the family. There is variation in the severity of DMD and individual life-expectancy.

www.musculardystrophy.org/aboutmusculardystrophy/conditions/97duchenne-musculardystrophy00

Juvenile Huntington's Chorea

This is 'a less common, early-onset form of Huntington disease that begins in childhood or adolescence and usually before 20 years of age. It is a progressive disorder that causes the breakdown of brain cells in certain areas of the brain. This results in uncontrolled movements, loss of intellectual abilities, and emotional disturbances. Juvenile Huntington disease has a rapid disease progression once symptoms present. There currently is no cure. Treatment is supportive and focused on increasing quality of life. Most people with juvenile HD do not survive more than 10 to 15 years after symptoms begin <https://rarediseases.info.nih.gov/diseases/10510/juvenile-huntington-disease> (Accessed 25th January 2019).

Rare life-limiting or life-threatening conditions

A rare disease is defined by the European Union as one that affects less than five in 10,000 of the general population. There are between 6,000 and 8,000 known rare

diseases and around five new rare diseases are described in medical literature each week. This equates to approximately 3.5 million people in the UK and 30 million people across Europe. In the UK, a single rare disease may affect up to about 30,000 people. The vast majority of rare diseases will affect far fewer than this. Some will affect only a handful, or even a single person, in the whole of the UK. 75% of rare diseases affect children. Rare diseases include rare cancers such as childhood cancers and some other well-known conditions, such as cystic fibrosis and Huntington's disease. At the time of completing this thesis, the Department of Health England is publishing a strategy to address the diagnosis, numbers and needs of children born who acquire rare conditions during childhood.

<https://www.raredisease.org.uk/what-is-a-rare-disease/>

Appendix J: Research prompts

Research aim

To gain a better understanding about the sexuality and relationship choices and experiences in young adults with life-limiting conditions, as well as how best to support their needs and those of the parents/carers and care staff who support them. Aiming to explore the experiences, perceptions, choices, legal issues and taboos about sexuality and reproduction of young adults with LLTCs and their supporters.

Young adults

- Your views about relationships, sex and reproduction?
- Whether you think you need any advice or training on some or all of these matters?
- What, if any, sex education you receive at school, college, online or after leaving school?
- Had a boyfriend/girlfriend/same sex relationship/both or no relationships?
- Would like children or not? Preparation for this (genetic counselling, fertility advice)?
- Considerations about the impact of the LLTC on sex and sexuality
- If you feel it would be helpful for your carers or staff to know more about sex and relationships for people with conditions like yours?

Partners/ Parents/carers

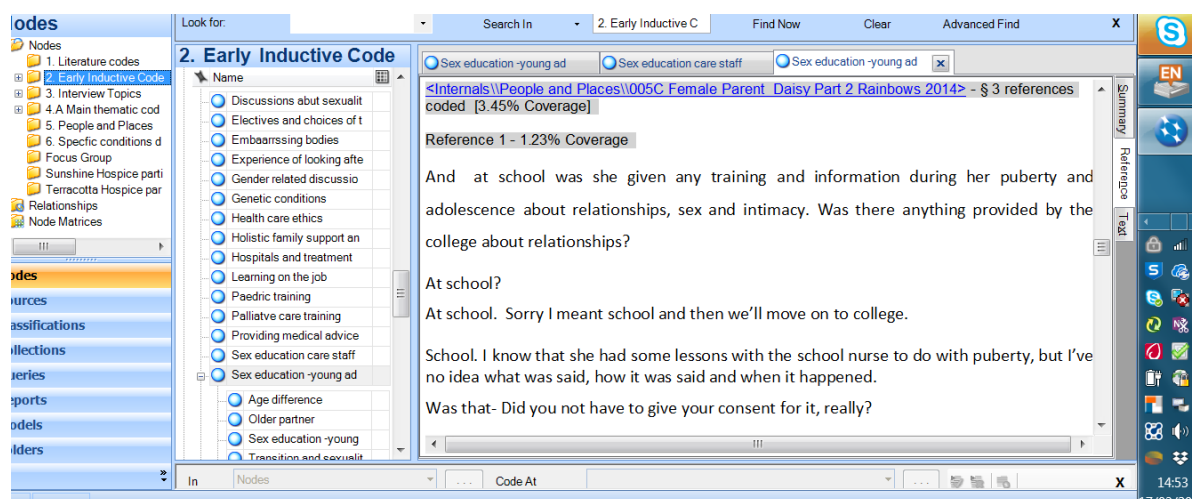
- Your views about relationships, sex and reproduction in people with life-limiting conditions?
- Whether you think either you or your son/daughter would benefit from advice or training on these matters?
- Whether your son/daughter had training on sex and relationship education and from whom and where?
- Whether your son or daughter is in/has had/or would like a relationship with opposite sex/same sex/not sure?
- Whether your son/daughter would like children and your views and preparation for this, such as genetic counselling, fertility advice?
- If you feel it would be helpful for other carers or staff to know more about sex, reproduction and relationships for people with life-limiting conditions?

Care staff

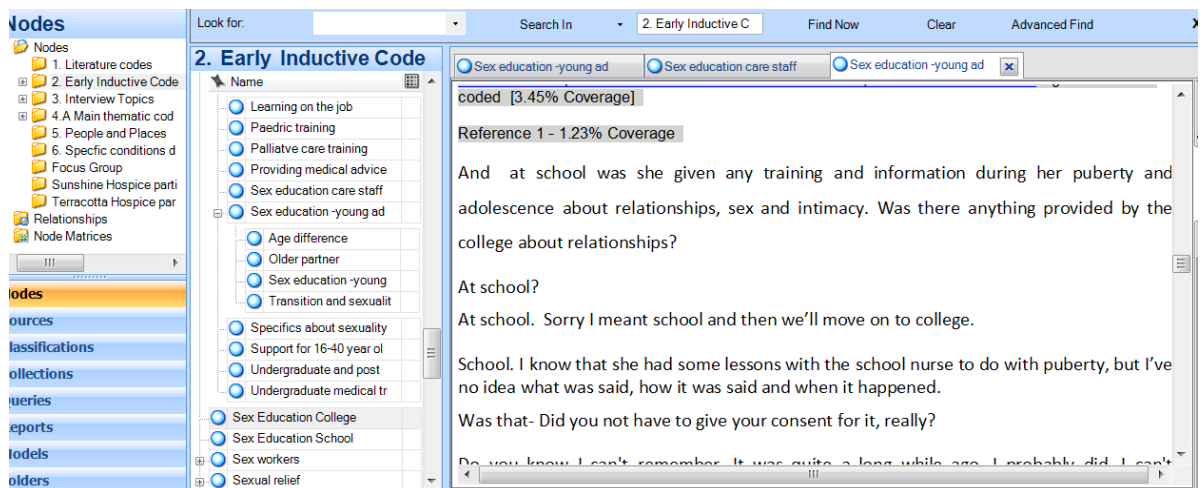
- Your views about intimacy, relationships, sex and reproduction for people with life-limiting conditions?
- Considerations about relationships, intimacy and sex?
- Whether you think care staff would benefit from advice or training on these matters?
- Received training on the sexuality of people with LLTCs?
- If you feel it would be helpful for parents and other carers to know more about sex, reproduction and relationships for people with life-limiting conditions?
- Views about reproduction on an uncertain life course?

Appendix K: An excerpt of an interview transcript, illustrating how it was analysed.

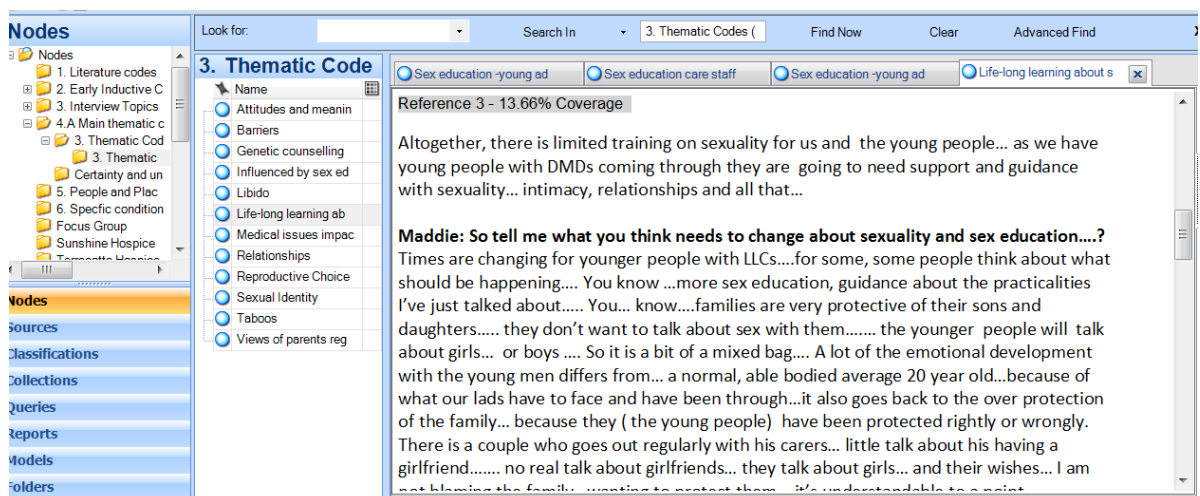
In the following excerpts from an interview transcript below, regarding access to sex education, I show the various stages of my coding and analysis.



Appendix K(i): In the above initial (early inductive) coding of the transcript, using NVivo 10 qualitative software, I identified a large number of codes related to sex education (SRE). Before coding, I read and re-read the transcripts and listened to the digital recordings to develop an initial long list of codes. A large number of codes emerged addressing various aspects of sex education in all four groups of participants.



Appendix K(ii): In the above, I then collapsed many of my initial codes into concepts/categories related to sex education. These were then organised to focus on themes, such as where sex education was received, the forms and types of learning available and access to specialist sex education.



Appendix K(iii): In the above, these were then organised to focus on themes related to sex education (across the four groups of participants'). The above is an excerpt from a care practitioner's transcript.

Nodes

- 1. Literature codes
- 2. Early Inductive Cod
- 3. Interview Topics
- 4. A Main thematic cod
 - 3. Thematic Codes
 - Certainty and uncer
 - 5. People and Places
 - 6. Specific conditions d
 - Focus Group
 - Sunshine Hospice part
 - Terracotta Hospice par
 - Relationships
 - Mede Matings

Nodes

Sources

Classifications

Collections

Queries

Reports

Models

Folders

Look for: Search In 3. Thematic Codes Find Now Clear Advanced Find

3. Thematic Code

Name

- ☐ Influenced by s
 - ☐ Negatively i
 - ☐ Positively inf
- ☐ Knowledge abo
- ☐ Media
- ☐ Sex education
- ☐ Sex education
- ☐ Sex education f
- ☐ Sex education f
- ☐ Sex education f
- ☐ Sex education f
- ☐ Sex education f
- ☐ Specialist sex e

☐ Medical issues imp

☐ Relationships

☐ Reproductive Choi

☐ Sexual Identity

☐ Taboos

Specialist sex education r

<Internals\People and Places\018 020714 Topaz Mother the Js and part two Vincent> - § 1
reference coded [0.75% Coverage]

Reference 1 - 0.75% Coverage

whether it's in a hospice or in the hospital or in the community or general practice, they should have better training on sex, relationships, intimacy and genetic screening? Or do you think they're just embarrassed raising some of those issues when they're trying to deal with a fairly complex condition?

Topaz You think, I think they would be embarrassed? Well, if they are they shouldn't be in that job should they?

No, no.

You know I mean that's, they're a service there to help people like Vincent.... whatever illness. No, training about sex, intimacy, relationships, genetic screening, IVF, that should be there definitely. And nobody should be embarrassed about it.

Appendix K(iv): An excerpt of transcript from a parent about sex education provision.